

**School of Occupational Therapy, Social Work and Speech Pathology**

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**Autism in regional and remote communities: Examining the effect of an  
autism spectrum disorder diagnosis on regional and remote families and  
innovative therapies**

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**This thesis is presented for the Degree of  
Doctor of Philosophy  
of  
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# Author's Declaration

To the best of my knowledge and belief, this thesis does not contain material previously published by any other person. This thesis does not contain material which has been accepted for the award of any other degree or diploma in any university. No other person's work has been used without due acknowledgement.

The research presented and reported in this thesis was conducted in accordance with the National Health and Medical Research Council National Statement on Ethical Conduct in Human Research (2007) – updated March 2014. Human research ethics approval for this research was received from the Curtin University Human Research Ethics Committee (EC00262), Approval Number: HR123/2014

Signature: \_\_\_\_\_

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# Statement of Contributors

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# Abstract

With the trend of increasing diagnosis and prevalence of autism spectrum disorder (ASD) in children, the need for feasible, effective and appropriate interventions for this group is also steadily growing. Families living in regional and remote areas in Western Australia (WA) experience many barriers, including having to travel considerable geographical distances to attend therapy appointments, limited screening leading to delayed diagnoses, and challenges in the retention of skilled health professionals in these areas. There is a need to develop innovative service models, methods and interventions to address these barriers for families of children with ASD; information communication technologies (ICT)-based interventions are one such innovation emerging as a viable delivery model for early intervention in this population. Preliminary research suggests ICT-based interventions have utility to ameliorate some of the challenges families of children with ASD experience; however, there remains a lack of rigorous research in relation to ICT's effectiveness and appropriateness for children with ASD. One promising ICT-based intervention is the Therapy Outcomes By You application (TOBY app). The TOBY app is an iPad application and intervention tool developed by a team of WA computer scientists, psychologists and speech pathologists. The TOBY app is based on early intensive behavioural intervention guidelines supporting high-intensity interventions to address individual children's needs using behavioural, educational and developmental approaches with a focus on the following four major skill areas in its syllabus: 1) visual motor; 2) imitation; 3) language; and 4) social. The TOBY app is delivered within a rigorous learning framework of stimulus adjustment relative to performance, with appropriate mastery criteria and prompting. The TOBY app presents a choice of tasks, drawn from a curriculum tree, based on the child's progression beyond pre-requisite tasks. The TOBY app is designed to provide a low cost and easily accessible intervention method to supplement existing therapy.

The TOBY app can be utilised by parents and their children with ASD without direct input from health professionals. Because the TOBY app can be completed in the homes of families under the guidance of caregivers, this novel intervention method holds significant promise for enabling families of children with ASD living in regional areas access to services they may otherwise have been unable to access. Using the United Kingdom Medical Research Council (UKMRC) guidelines for the development and evaluation of complex interventions, the overall aim of this PhD project was to examine the feasibility, effectiveness and appropriateness of the TOBY app for children with ASD living in regional and remote WA. The UKMRC guidelines comprise four phases: 1) the development phase, where the components of the interventions are defined; 2) the feasibility or piloting phase, where the trial and intervention design are defined, including appropriateness and feasibility; 3) the evaluation phase, involving the use of an RCT to evaluate effectiveness; and 4) the implementation phase, where the effective long-term implementation of the intervention into practice is completed, including long-term follow-up. These four phases can either be executed using a linear sequential process or in a more iterative fashion, moving backwards and forwards throughout the development and evaluation process.

The overall aim of the thesis was addressed in five individual studies presented as peer-reviewed journal manuscripts: 1) a systematic review; 2) a cross-sectional survey; 3) an intervention study (exploratory randomised controlled trial); 4) semi-structured interviews with parents who used the TOBY app within the intervention study; and 5) a long-term follow-up study with participants. These five papers are structured in three distinct phases within the thesis. Firstly, Chapters 2 and 3 aim to contextualise the factors within which the TOBY app were evaluated and implemented, a key aspect in the design and evaluation of complex interventions outlined in the UKMRC guidelines. Chapter 2 comprises of a

systematic review of existing literature for parent-mediated interventions delivered remotely for families living outside of regional areas. Chapter 3 reports on findings from a study using a cross-sectional survey aimed to explore the impact of regionality on stress levels, coping, quality of life (QoL) and daily routines for parents and families of a child with ASD (aged 2-18 years) in WA. Chapter 4 details the second phase — the evaluation phase of the UKMRC — whereby the effectiveness of the TOBY app was evaluated using an exploratory randomised controlled trial with families living in regional areas. The third phase is comprised by Chapters 5 and 6, the implementation phase of the UKMRC framework, which consists of two studies involving in-depth interviews and a long-term follow-up with participants from the effectiveness study were conducted.

Chapter 2 synthesises the existing literature for parent-mediated interventions delivered remotely to families living outside of major city areas who have a child with ASD using a systematic review. The preferred reporting items for systematic reviews and meta-analyses (PRISMA) statement was used to guide the methodology and report the findings of this study. Five electronic databases were comprehensively searched for studies investigating parent-mediated interventions delivered remotely for families living outside of major city areas who had a child with ASD. Two independent researchers reviewed the articles for inclusion, and assessment of methodological rigour was based on the Kmet appraisal checklist. Key findings from Chapter 2 were that there is preliminary evidence for parent-mediated intervention training delivered remotely, to improve parent knowledge, increase parent intervention fidelity, and improve the social behaviour and communication skills for children with ASD. However, due to the low number of RCTs, difficulty in defining the locality of the population and a paucity in using standardised outcome measures, generalisation of the findings to the target population are limited. Key recommendations for future research derived from Chapter 2 were the need for: 1) researchers to use standard

classifications systems when describing their participants living in regional and remote areas to improve the applicability and generalisation of findings for the reader; 2) further investigation into the unique context of families of children with ASD living in regional and remote settings; and 3) further investigation into parental experiences in using parent-mediated interventions to assess their appropriateness. Findings from Chapter 2, in conjunction with existing literature for ICT-based interventions, informed the next stages in the thesis.

The aim of Chapter 3 was derived from a key recommendation in Chapter 2 — the need for further investigation into the unique context of families of children with ASD living in a regional and remote setting. Chapter 3 details findings from a study using a cross-sectional survey aiming to explore whether regionality is associated with differing stress levels, coping, quality of life (QoL) and daily routines for parents and families of a child with ASD (aged 2-18 years old) in WA. Further, the study aimed to compare the stress, coping, QoL, and daily routines of parents of children with ASD living in WA to the general population using Australian normative data. A sample of 278 families were recruited into the study from WA. All families had a child or adolescent (2-18 years old) with ASD. Findings suggested that despite having higher levels of stress and lower QoL compared to the general population, residing in a geographically low densely populated area in WA was associated with a higher preference for avoidant coping styles, and has no association on stress levels, QoL or daily routines for parents who have children with ASD.

Chapter 4 is comprised of an exploratory waitlist parallel RCT study to evaluate the feasibility and effectiveness of the TOBY app for families living in regional WA. Fifty-nine children with ASD and their families were randomised into either the therapy-as-usual or intervention group. With the exception of expressive language, no significant between-

group differences were detected for any of the outcomes assessed. When all participants who received the intervention were pooled (intervention and control group) and measured over time, improvements were shown in receptive and pragmatic language and social skills; these gains were maintained at three months post-intervention, thus suggesting skill acquisition. The key findings from Chapter 4 were that when compared to therapy-as-usual, the TOBY app provided benefit in improving the expressive language for children with ASD living in regional areas of WA; and that from a feasibility perspective, adherence and intervention fidelity were poor, indicating a need to develop and implement evidence-based strategies aimed at improving adherence to ICT-based interventions for parents and children with ASD.

The study outlined in Chapter 5 aimed to evaluate the appropriateness of the TOBY app intervention from the perspective of 24 parents who participated in the effectiveness trial of the TOBY app (Chapter 4). Data from parent interviews were analysed using a thematic analysis approach. Themes generated were mapped against a theoretical framework to evaluate the appropriateness of the intervention to determine the impact of the TOBY app from the perspective of the parents who participated in the RCT. When the identified themes were mapped against an appropriateness framework, the key findings from Chapter 5 were that parents were of the opinion that the TOBY app was relevant and important to them and their children's needs, while expressing partial support for using the TOBY app as: 1) a positive experience for them and their children; 2) beneficial for them and their children; 3) a socially and ecological valid intervention; and 4) an intervention that supported change and continuation in the skills learnt.

Chapter 6 comprises a follow-up study with 15 participants from the RCT discussed in Chapter 4. The study aimed to determine if these 15 participants maintained or continued to

improve their language and social communication skills at least 12 months after using the TOBY app in the RCT. The study also aimed to explain the maintenance of skills and ongoing use of the TOBY app from the perspectives of the parents who participated in the RCT. Findings suggested the children with ASD maintained their improved skills in expressive language at least 12 months post-intervention. However, no significant skill improvements were detected for symbolic play, visual motor, fine motor or expressive language.

Guided by the UKMRC framework, the findings from the three phases of research in this thesis demonstrated that the TOBY app is an effective, feasible and appropriate intervention for improving the developmental skill of expressive language, with low-level evidence supporting its effectiveness in improving receptive language skills, pragmatic language skills, and social communication skills. Implications from the findings in this thesis highlight the importance for researchers to better define the participants from regional and remote areas in their studies to improve the interpretation and generalisation of research results to the relevant population. Moreover, findings indicate that ICT-based interventions are just one tool to address the challenges experienced by families with children with ASD. Parents reported that ICT-based interventions cannot, and should not, replace face-to-face instruction and that ongoing support by trained therapy professionals is required. Lastly, researchers and developers of ICT-based interventions for children with ASD could increase the likelihood of the adoption of their interventions through an increased understanding of the Diffusion of Innovation theory, to not only improve intervention fidelity and adherence, but to ensure widespread use within the ASD community. Future research for ICT-based interventions includes the following recommendations: 1) capturing mental health issues, in addition to experiences of stress and QoL for parents of children with ASD living in regional and remote areas; 2) targeted



recruitment of more participants from remote and very remote areas in effectiveness trials for ICT-based interventions; 3) improved understanding into the negative or adverse effects of using ICT-based interventions with children with ASD; and 4) evaluation of the cost-effectiveness of ICT-based interventions for children with ASD.



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# Dedication

To my girls Eve and Alice, stay awesome!

Dad



# List of Publications

This doctoral thesis consists of the following publications:

1. Parsons, D., Cordier, R., Vaz, S., & Lee, H. C. (2017). Parent-mediated intervention training delivered remotely for children with Autism Spectrum Disorder living outside of urban areas: Systematic review. *Journal of Medical Internet Research*, 19(8), e198. doi:10.2196/jmir.6651
2. Parsons, D., Cordier, R., Lee, H., Falkmer, T., & Vaz, S. (In Press). Stress, coping, and quality of life in families with a child with ASD living regionally. *Journal of Child and Family Studies*.
3. Parsons, D., Cordier, R., Lee, H., Falkmer, T., & Vaz, S. (2019). A randomised controlled trial of an information communication technology delivered intervention for children with autism spectrum disorder living in regional Australia. *Journal of Autism and Developmental Disorders*, 49(2), 569-581. doi:10.1007/s10803-018-3734-3
4. Parsons, D., Wilson, N. J., Vaz, S., Lee, H., & Cordier, R. (2019). Appropriateness of the TOBY application, an iPad intervention for children with Autism Spectrum Disorder: A thematic approach. *Journal of Autism and Developmental Disorders*, Online. doi:10.1007/s10803-019-04115-9
5. Parsons, D., Vaz, S., Lee, H., Robinson, C., & Cordier, R. (Under review). A twelve-month follow-up of an information communication technology delivered intervention for children with autism spectrum disorder living in regional Australia. *Journal of Autism and Developmental Disorders*.

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# List of Abbreviations

ABA: Applied Behaviour Analysis

ABS: Australian Bureau of Statistics

ASD: Autism Spectrum Disorder

ASGC: Australian Standard Geographical Classification

App: iPad<sup>®</sup> Application.

CONSORT: Consolidated Standards of Reporting Trials

DSM: Diagnostic and Statistical Manual of Mental Disorders

EIBI: Early Intensive Behavioural Interventions

ICT: Information Communication Technology

QoL: Quality of Life

RCT: Randomised Controlled Trial

SEIFA: Social-Economic Indexes for Areas

SD: Standard Deviation

TOBY: Therapy Outcomes By You

UKMRC: United Kingdom Medical Research Council

WA: Western Australia



# Explanation of Terms

Throughout this thesis, the term information communication technology (ICT) has been used to describe the method of intervention delivery. There is no universal consensus on the terminology when describing using technology and in relation to delivering interventions to children with autism spectrum disorders, so I have chosen to use the term ICT for consistency and ease of reading. The term ICT includes but is not restricted to computer-based, online, web-based, tablet, and information technology (IT) methods of intervention delivery.

As most children who participated in my research (except for the those in Chapter 3) were diagnosed under the new Diagnostic and Statistical Manual of Mental Disorders (DSM-5) (American Psychiatric Association, 2013), I chose to use the term Autistic Spectrum Disorder (ASD) to describe Autism. The term is inclusive of people with a diagnosis of Asperger's Syndrome and Pervasive Developmental Disorder – Not Otherwise Specified as described in the DSM-IV.

In my thesis, I have adopted the terms to classify and describe regionality based on the Australian Standard Geographical Classification (ASGC), the classification system used by the Australian Bureau of Statistics (ABS) for the collection and dissemination of geographically classified statistics (Australian Bureau of Statistics, 2003, 2011). The ASGC remoteness structure is classified into five remoteness areas (RAs) and include the following categories: 1) major cities; 2) inner regional; 3) outer regional; 4) remote; and 5) very remote. RAs are based on road distances to the nearest service centres and average scores are calculated using the Accessibility/Remoteness Index of Australia (ARIA+) grid, which is a one square kilometre grid covering all of Australia (Australian Bureau of Statistics, 2011; McAuliffe, Vaz, Falkmer, & Cordier, 2016). I have chosen to use the term

regional and remote throughout the majority of my thesis to describe individuals and families living outside of major cities areas. The only exception to this nomenclature was the use of the term ‘non-urban’ in Chapter 2, which was used to refer to regional and remote areas. As Chapter 2 had been published as a manuscript at the time of writing this thesis, I was unable to change the terminology to maintain consistency throughout the thesis. In Chapter 3, I used the terms low densely populated (LDP) and densely populated (DP) areas. Both LDP and DP areas comprise of RAs from the ASGC; hence, additional terms were generated as no terminology for the RAs existed within the ASGC system. In this thesis, LDP areas comprise the very remote, remote and outer regional RAs, whereas inner regional and major cities RAs were grouped as DP areas.

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# Chapter 1 Introduction

## 1.1 Background to research

The impetus for this research was the culmination of opportunity and my personal experiences and research interests. The need and, subsequently, the opportunity for this project stemmed from years of negotiations with stakeholders who anecdotally reported a dearth of early intervention services for children with autism spectrum disorder (ASD) in southwest Western Australia (WA). In 2014, the School of Occupational Therapy, Social Work and Speech Pathology were approached by the Lishman Health Foundation, to undertake a staged study to help understand the experience of families with a child with ASD and test the effectiveness of an early intervention iPad application, the Therapy Outcomes By You application (TOBY app) that is designed to complement routine therapy. The Lishman Health Foundation is an independent charitable organisation that funds, facilitates and promotes health research that has significant health gain for individuals, families and communities living in south-west WA and, more broadly, regional Australia.

I spent the early years of my life in the southwest of WA where this research took place. Towns in the southwest of WA are filled with a strong sense of community connection and spirit. Having grown up in the southwest of WA, I have a natural affinity for the community and a keen interest in helping fill the knowledge gap to improve the lives of people who live there. Further, technology has always interested me. I am of the view that the use of Information Communication Technology (ICT)-based interventions hold significant promise in improving health and community outcomes, particularly for people living in regional areas. The nexus of these aspects of the study strongly drew me into my PhD journey and maintained the momentum throughout the impending ups and downs.

Throughout the research, my interest in the individual lives of these families and their diverse experiences in having access to services and their views on the quality of services continued to evolve. Individual anecdotes and accounts of restricted access to services highlight the unique experience that families living in regional and remote areas experience when trying to achieve the best outcomes for their children. These experiences were juxtaposed with my own lived experience of having two delightful little girls. Watching them grow, develop, connect and understand their world is an incredible journey, and having this experience in parallel with the families from my research, who were often struggling with their parenting journeys, has forever impacted my personal and professional journey; motivating me to improve the collective knowledge within this area. What struck me most, though, was the absolute commitment by all parents to help their children to achieve the best possible life they can; the resilience and unique community spirit were evident in all families involved in this research.

## **1.2 Context**

My research is not the only study conducted to investigate the effectiveness of the TOBY app. The randomised controlled trial (Chapter 4) in regional areas ran parallel to a multi-centre trial conducted by Whitehouse et al. (2017) with a population primarily from major cities (Perth, WA and Melbourne, Victoria). As this PhD project ran in parallel with the study conducted by Whitehouse et al. (2017), the standardised measures and methodologies were similar for both studies. This was to enable comparison between the major city group and regional groups in relation to the intervention. While comparisons will be made between the Whitehouse et al. (2017) RCT and my RCT study, all the processes, procedures and findings resulting from this research project, as described in this thesis, are my unique contribution to the knowledge base. Furthermore, the findings from the Whitehouse et al. (2017) had not been published by the time I commenced data collection

for my RCT; hence, their findings did not inform my research.

Within this introductory chapter, I will briefly discuss ASD and provide an overview of the WA context within which my research was undertaken. Next, I will describe early intensive behavioural intervention (EIBI) and applied behaviour analysis (ABA), with specific reference to innovations that have occurred to the delivery of EIBI in recent times, and in particular, to ICT. Following this, I will describe the theoretical frameworks that I have used to guide my research and some of my recommendations for future research and implications for clinical practice. Lastly, I will conclude this chapter with an outline of this thesis and overall aims.

### **1.3 Raising a child with ASD in regional Australia**

#### **1.3.1 ASD**

ASD is a multidimensional and pervasive neurodevelopmental condition characterised by impairments in social communication and social behaviour including: difficulty developing age-appropriate friendships; difficulty with interpreting non-verbal gestures; adherence to rigid routines, interests or activities; and high sensitivity to changes in their environment (American Psychiatric Association, 2013; Happé & Ronald, 2008). Prevalence rates of ASD reported in the literature vary from 20 per 10,000 to as high as 110 per 10,000 of the global population of ASD (Baio, 2012; Baird et al., 2006; Baron-Cohen et al., 2009; Hill, Zuckerman, & Fombonne, 2015; Kim et al., 2011; Kogan et al., 2009; Matson & Kozlowski, 2011; Williams, MacDermott, Ridley, Glasson, & Wray, 2008). A consistent trend has been observed in recent times, with significant increases in the prevalence of ASD in children being reported. The literature suggests a number of plausible explanations for this trend, including: earlier diagnosis; increased awareness of ASD in the community; and updates to the diagnostic criteria for ASD, specifically the broadening of the diagnostic

criteria in the Diagnostic and Statistical Manual of Mental Disorders (DSM-5) (American Psychiatric Association, 2013; Matson, Wilkins, Boisjoli, & Smith, 2008).

Regardless of the aetiology, the increasing prevalence of ASD exerts significant demands on early intervention services and, as a result, health and education institutions could benefit from innovative service delivery models and methods to address the increased burden on resources. This is no more pertinent than in the case of families of children with ASD living in regional and remote areas, where barriers to access needed services are inflated due to considerable geographical distances to attend therapy appointments (Dew et al., 2013; Dew et al., 2012).

### **1.3.2 Regional and Remote Australia**

In my preliminary exploration of literature related to ASD, it quickly became apparent that despite considerable research into ASD, there was a lack of focus on families of children with ASD specifically living in regional and remote areas. The unique experiences of families living in this context were scarcely represented, with limited published research on this population lacking detail and consistency when describing their participants. Further, it became apparent from the limited literature that families of children with ASD living in regional and remote areas are a unique population. While similarities may exist between families of children with ASD in major cities and families of children with ASD in regional and remote areas, there are a number distinctive characteristics in families with children with ASD living in regional and remote areas due to barriers these families experience as a consequence of their geographical remoteness (Hoogsteen & Woodgate, 2013b). To my knowledge, there has been no research conducted into the unique experiences of these families living in regional and remote WA.

WA has some distinct demographic, geographical and institutional characteristics that

warrant explanation to aid in contextualising my research, particularly for audiences unfamiliar with Australia. As the largest state in Australia, WA comprises a total landmass of 252,641,768 hectares, approximately 30% of the total size of Australia (Australian Bureau of Statistics, 2017). Despite the large landmass, the population of WA is 2.59 million people; only 10% of the total population of Australia. Notably, approximately 80% of people in WA live in the capital city, Perth. This makes WA the least densely populated state in one of the least densely populated countries in the world (three people per km<sup>2</sup>) (Australian Bureau of Statistics, 2017). For the sake of comparison, Canada has a similar population density (four people per km<sup>2</sup>), while the United Kingdom has considerably more (274 people per km<sup>2</sup>; Office of National Statistics, 2019; Statistics Canada, 2015). The low population density presents challenges for the provision of healthcare and disability services for individuals and families living in regional and remote areas of WA (Dew et al., 2013; Dew et al., 2012).

Health services in Australia are delivered via a complex system of funding and governance structures. The Commonwealth of Australia (henceforth referred to as the Federal Government) and State/Territory Governments provide the vast majority of services, which are augmented with privately operated service providers. Aside from funding most of the health services in Australia, the Federal Government provides leadership in the areas of national health policies and research. The Federal Government funds and administers the national Medicare Benefits Schedule, Pharmaceutical Benefits Scheme and the National Disability Insurance Scheme—critical services in the context of Australian primary health care and disability service provision. Furthermore, the Federal Government primarily funds residential aged care sectors, veteran services, and primary health care for Aboriginal Australians. State and Territory governments are responsible for the management and delivery of acute and sub-acute hospital health services, psychiatric hospital care services

and a myriad of other health services such as school health, dental health, and, importantly in the context of this thesis, community-based child and maternal health services (Attorney-General's Department, 2008).

In regional and remote Australia, the State and Territory Governments have responsibility for delivering the vast majority of health services through rural health and hospital services. One-third of Australians do not live in major cities; however, the proportion of primary health services providers in regional and remote areas does not reflect the population distribution (Australian Bureau of Statistics, 2017; Dew et al., 2012). Specifically, there is a recognised shortage of adequately trained health professionals, including general practitioners, dentists, pharmacists and allied health professionals in communities living in regional and remote Australia; with decreasing services broadly proportionate to the distance away from major city centres (Australian Bureau of Statistics, 2011; McGrail & Humphreys, 2009). Additionally, knowledge and skills in managing developmental disabilities, including ASD, in regional and remote areas is lacking, with retention and poor consistency of adequately trained health professionals in these areas noted as a critical contributor (Dew et al., 2012).

A 2014 study estimated that, on average, the financial cost of having a child with ASD in WA for families is AU\$34,900 per annum (Horlin, Falkmer, Parsons, Albrecht, & Falkmer, 2014). The study reported that the highest cost was due to a loss of income accounting for approximately 90% of the total cost (AU\$29,200) as a result of being unable to work. Out-of-pocket treatment costs account for approximately AU\$4,800. Importantly for families living in regional Australia, the impact of travelling increased distances to appointments or other services for their child, compared with their city-dwelling counterparts, could elevate the financial burden given the increased travel costs and impact on their employment. The

impact is further exacerbated by a paucity of flexible employment opportunities in these communities. The researchers also reported a link between the financial burden and ASD symptom severity, suggesting that EIBI addressing core ASD symptom severity could lead to decreased financial costs (Horlin et al., 2014).

My initial exploration of current literature supported anecdotal reports that families of children with ASD living in regional and remote areas commonly experience several barriers to improving the outcomes for their child. These barriers include a delay in diagnosis due to reduced screening programs and considerable travel distances to access suitably qualified clinicians for evidence-based therapy services (Hutton & Caron, 2005; Murphy & Ruble, 2012; Smith, Humphreys, & Wilson, 2008; Struber, 2004). Further, parents of children with ASD living in regional and remote areas who may experience increased financial pressures are often forced to undertake additional responsibilities related to the health care of their children and are at high risk of feeling isolated on how best to support their children with ASD (Hoogsteen & Woodgate, 2013a). To date, no research has been conducted into the coping strategies, stress levels, quality of life (QoL) or daily routines for parents living in regional and remote WA.

### **1.3.3 Early Intensive Behavioural Intervention**

Much of the research on interventions for ASD stresses the importance of EIBI (Howlin, Magiati, Charman, & MacLean, 2009). Common EIBI interventions cited in the literature include the Early Start Denver Model, ABA, Discrete Trial Training, and Pivotal Response Training (Dawson et al., 2010; Prior & Roberts, 2012; Thomson, Martin, Arnal, Fazzio, & Yu, 2009). All of these interventions require considerable therapy time (in the form of highly trained personnel) and financial resources (Whalen, Liden, Ingersoll, Dallaire, & Liden, 2006). For example, the Early Start Denver model requires the participants to

complete four hours of intervention, five days per week over a two-year period (Dawson et al., 2010).

With such a high amount of resources required to implement EIBI effectively, innovative models of service delivery for interventions, such as parent-mediated, telehealth or ICT-based delivered interventions may hold the key to augment current services. These type of delivery methods may have the potential to decrease access barriers and increase dosage for children with ASD, while not compromising effective intervention delivery, particularly for families living in regional and remote communities.

#### **1.3.4 Applied behaviour analysis**

ABA is accepted as one of the most effective treatments for children with ASD, often being cited in clinical guidelines for EIBI for children with ASD (Prior & Roberts, 2012; Wilczynski et al., 2009). ABA uses techniques to improve language skills (receptive and expressive), adaptive behaviour skills (social communication skills), motor skills, daily living skills and reduce problem behaviours often associated with ASD such as tantrums, self-injury, non-compliance, self-stimulation, and aggression (Heitzman-Powell, Buzhardt, Rusinko, & Miller, 2014; Virués-Ortega, 2010). ABA is not a specific program per se, but a set of principles and processes to identify which skills to improve and which problem behaviours to decrease. Further, ABA principles aim to identify modifications to the environment to facilitate ways to support and maintain positive skill and behaviour changes (Heitzman-Powell et al., 2014).

ABA uses operant conditioning to improve the child's skills (Venkatesh, Phung, Duong, Greenhill, & Adams, 2013). Children are presented with stimuli and if the desired behaviour is elicited, then positive reinforcement is provided to the child in the form of a reward or verbal praise. Prompting of the desired behaviour by the facilitator may be



required if the child does not exhibit the desired behaviour. Over time, the goal is to reduce (fade) the prompt (and reinforcements) to a point whereby the child self-initiates the desired behaviour. Throughout the process, the behaviour of the child needs to be measured to decide: 1) if a behaviour is desirable, when should it be reinforced; 2) when to prompt and the required magnitude of the prompt; 3) when a skill is mastered; and 4) what skills to work on next once a skill is mastered (Baer, Wolf, & Risley, 1968; Venkatesh et al., 2013).

Although ABA has demonstrated effectiveness for children with ASD, it is expensive and time-consuming to implement in both the short- and long-term. Furthermore, the delivery of ABA requires formal training that is often reserved for clinicians working towards becoming a licensed ABA practitioner (Heitzman-Powell et al., 2014). For families living in regional and remote Australia, parents or caregivers may be required to play a larger role when compared to their major city-dwelling counterparts in the provision of therapeutic services for their children with ASD to help overcome service access barriers (Dew et al., 2013; Hoogsteen & Woodgate, 2013b).

While the ABA programs are primarily designed and delivered by qualified practitioners, given the large recommended dosage, parents of the children with ASD are often trained to deliver ABA-based programs in the home environment. With the appropriate training and ongoing guidance, parents can become active agents in the therapeutic process, thereby delivering these interventions to their children in a more consistent manner (McConachie & Diggle, 2007). The inherent characteristics of ABA, such as its structured progressions, measurement-taking and storage, and stimulus-based learning make it an ideal fit for adaption to ICT-based delivery (Venkatesh et al., 2013).

## **1.4 Innovation in ASD interventions**

### **1.4.1 Parent-mediated and telehealth interventions**

The methods of delivery for EIBI for children with ASD are constantly evolving through innovation. Parent-mediated and telehealth interventions are two of the more prevalent and researched service delivery methods for children with ASD. Several systematic literature reviews have been conducted investigating parent-mediated interventions with findings providing some support for the use of parent-mediated interventions in children with ASD improving language, social behaviour and parent-child relationships (French & Kennedy, 2018; Green & Garg, 2018). However, findings are largely inconclusive and inconsistent due to weak research designs, resulting in an inability to draw strong conclusions (Diggle & McConachie, 2002; McConachie & Diggle, 2007; McConachie & Oono, 2013; Meadan & Daczewitz, 2015). Telehealth interventions have also received support in the literature to provide education, guidance, supervision, training, and coaching for the implementation of behavioural interventions sessions to parents or caregivers with children with ASD, as well as the delivery of behavioural and diagnostic assessments (Boisvert, Lang, Andrianopoulos, & Boscardin, 2010; Wainer, 2014). Both intervention delivery methods show promise in the ability of parents becoming active agents in the delivery of EIBI to their children.

Importantly, no systematic review investigating the effectiveness of parent-mediated interventions for families with a child with ASD living in regional and remote areas has been conducted. While evidence for the effectiveness of these parent-mediated interventions is still developing, current findings suggest parent-mediated interventions demonstrate promise as alternatives to more traditional methods of service delivery, such as face-to-face clinician-delivered models, which could be useful for families of children

living in regional and remote Australia (Ingersoll & Berger, 2015; McConachie & Oono, 2013; Vismara, McCormick, Young, Nadhan, & Monlux, 2013). Moreover, these interventions demonstrate a clear need for researchers and developers to create innovative models of intervention delivery to address the exhausting resources required for more traditional models of service delivery. One of the best supported evidence-based EIBI interventions where parents can be active agents in the delivery of the intervention for children with ASD is ABA (Heitzman-Powell et al., 2014). A model of delivering parent-mediated interventions that is emerging as a viable option for families living in regional and remote areas is the use of ICT-based interventions.

## **1.5 Information Communication Technology-based interventions**

### **1.5.1 Proliferation of ICT**

With considerable technological advances in ICT across most of the world, alternative models of service delivery using ICT for health interventions have proliferated. ICT-based interventions have demonstrated high efficacy in a variety of areas in health care, such as delivering behavioural treatments for people with depression and anxiety, and there is emerging evidence of ICT-based EIBI for children with ASD (Ramdoss et al., 2011; Ramdoss et al., 2012; Reger & Gahm, 2009; Spek et al., 2007). Furthermore, ICT-based interventions can be tailored to suit the unique needs of children with ASD as these children tend to have a high preference for ICT given the user's ability to control the pace and the modality's high amount of structure, predictability, and asynchronous characteristics (Ploog et al., 2013; Rajendran, 2013). These aspects make ICT an ideal conduit to deliver interventions to children with ASD. In addition, ICT-based interventions have the potential to improve access to the appropriate therapy opportunities for those families who may, for a variety of reasons including cost, distance from services, and employment or family commitments, be unable to receive EIBI for their child. Given the

sustained and intense therapy required to deliver EIBI, ICT-based interventions are an ideal service delivery method for families living in regional and remote areas (Dew et al., 2013; Neely, Rispoli, Camargo, Davis, & Boles, 2013; Ploog, Scharf, Nelson, & Brooks, 2013).

Despite ICT-based interventions having the potential to improve access to EIBI for children with ASD, it should be noted that ICT-based interventions present the following unique challenges for this population: 1) parents may have limited financial and time resources to implement the interventions in the home environment; 2) the complexity involved in creating a design for software applications that are suitable for all children with ASD; and 3) poor generalisation from ICT-based intervention to “real world” situations (Ingersoll & Berger, 2015; Ploog et al., 2013; Silver & Oakes, 2001; Tseng & Do, 2010). Additionally, the characteristics of ICT-based interventions that make them a promising EIBI tool may compound existing challenges often experienced by children with ASD such as: 1) decreasing the interaction opportunities between the child with ASD and the person delivering the intervention, for example teachers, parents, or therapists; 2) increasing social isolation due to a reduced opportunity to practice social interactions; and 3) increasing the intense fixations on the ICT device, due to repetitive and restrictive behaviours displayed by many children with ASD (Ramdoss et al., 2011).

Evidence for ICT’s utility for children with ASD is in its early stages, with preliminary findings suggesting ICT has potential benefits in the diagnosis and delivery of interventions for this population (Boisvert et al., 2010; Boucenna et al., 2014; Doenyas, Şimdi, Özcan, Çataltepe, & Birkan, 2014; Falloon & Khoo, 2014; Golan & Baron-Cohen, 2006; McDuffie et al., 2013; Ramdoss et al., 2011; Ramdoss et al., 2012; Tanaka et al., 2010; Terry, 2009; Whalen et al., 2010). Despite these encouraging results, there is still a significant need for more research into the efficacy and feasibility of ICT-based

interventions for children with ASD. Moreover, there is a need to investigate the effectiveness of ICT-based interventions with different sub-groups in the ASD community, such as families living in regional and remote areas. Moreover, given the heterogeneous characteristics between populations, and the varying factors influencing the adoption and acceptance of these innovations as viable alternatives to more traditional interventions, the need is more pronounced in families with children with ASD.

### **1.5.2 Therapy Outcomes By You Application**

Designed by a team of WA computer scientists, psychologists and speech pathologists, the TOBY app is an ICT-based (iPad application) intervention designed to provide EIBI to children with ASD (Venkatesh et al., 2013). For families of children with ASD living in regional and remote WA who may experience difficulties in accessing therapy services, the TOBY app offers an innovative, cost-effective way to augment current services aimed at reducing impairments in social communication and social behaviour. The TOBY app uses ABA principles and is based on EIBI guidelines supporting high-intensity interventions to address individual children's needs using behavioural, educational and developmental approaches with a focus on the following four major skill areas in its syllabus: 1) visual motor; 2) imitation; 3) language; and 4) social. The TOBY app is delivered within a learning framework of stimulus adjustment relative to performance, with appropriate mastery criteria and prompting. The TOBY app presents children and their parents with a choice of tasks, drawn from a curriculum tree, based on the children's progression with pre-requisite tasks.

The TOBY app has three methods for therapy delivery: solo; partner; and Natural Environment Training (NET) (Venkatesh et al., 2013). The solo and partner activities are not new in ICT therapy intervention for this population (Ramdoss et al., 2011), however,

the NET activities are the most novel aspect of the TOBY app (Venkatesh et al., 2013). These activities aim to generalise the “on app” learning into natural situations by educating, prompting and logging parents’ translational intervention with their children. This generalisation to the natural environment has long been a weakness in ICT intervention for this population (Fletcher-Watson et al., 2016; Silver & Oakes, 2001). This component of the TOBY app addresses the issue around generalisation from software applications to ‘*real world*’ that other earlier generation computer-based interventions were lacking when attempting to provide therapeutic interventions to children with ASD (Hopkins et al., 2011; Ploog et al., 2013).

The TOBY app is designed not to replace one-on-one therapy with clinicians, but rather to provide a low cost and easily accessible intervention method to supplement existing therapy. Educating and training the parent to deliver some of the intervention is, according to its developers, the strength of the TOBY app. That is, the TOBY app has a focus on ‘*teaching a parent how to teach*’ (Venkatesh et al., 2013). Further, while not designed to be a substitute for face-to-face therapy, the TOBY app can be utilised by parents and their children with ASD without direct input from health professionals. Because the TOBY app can be completed in the homes of families under the guidance of caregivers, this novel intervention method holds significant promise for enabling regional and remote families to gain access to services they may have otherwise missed out on and increase the dosage of therapy the child receives (Moore et al., 2015; Venkatesh et al., 2013).

At the time of commencing my research, the TOBY app had been piloted in a study by Moore et al. (2015). In their study, Moore et al. (2015) provided the TOBY app to 33 participants aged 16 years or younger with a diagnosis of ASD recruited from a major city in Australia. Participants were asked to engage with the TOBY app for four to six weeks

without further coaching or promoting. Data were collected and analysed using descriptive statistics on usage patterns such as total time engaged in each of the three methods of delivery (solo, partner, NET), the number of sessions, completed learning units and the child correct/incorrect response to learning stimuli (Moore et al., 2015).

The findings from the study reported the participants engaged with the TOBY app tasks on average of 178.5 min (range 16.9-671.1 min) with only 23 out of 33 participants completing more than 100 learning units (range 0 to 4,182 units). The authors also reported a large variation in the proportion of the TOBY curriculum the participants completed, ranging from 17% to 100% of the total curriculum. Overall, the pilot trial concluded that the TOBY app might make a useful contribution to EIBI programming for children with ASD by delivering high rates of appropriate learning opportunities. Importantly for my research, the authors reported that even in the absence of therapist support, families were able to utilise the tool and, although usage patterns varied greatly, some families engaged with the TOBY app extensively and with good effect (Moore et al., 2015). Prior to my thesis, the TOBY app had not been evaluated for feasibility for children with ASD who were living in regional and remote areas. However, given the preliminary findings suggested that participants could use the TOBY app without further coaching, it was deemed a suitable intervention for this population.

## **1.6 Theoretical frameworks**

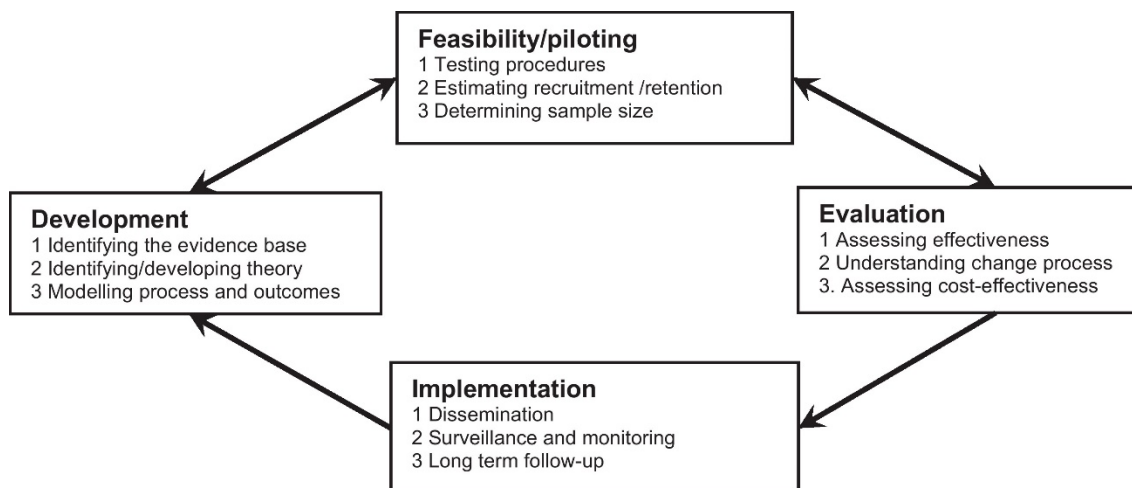
### **1.6.1 Design and Evaluation of Complex Interventions Development Framework**

In the early stages of undertaking this research, it became evident that I needed an overarching framework to guide me through the process. A robust, evidence-based process was required to maximise the methodological rigour of my research, given my inexperience as a researcher attempting to evaluate an innovative intervention with a

population that is underreported in the literature. Through my reading, I decided to use the seminal work by Campbell et al. (2000) in conjunction with the update by Craig et al. (2013) — the widely accepted framework developed by the United Kingdom Medical Research Council (UKMRC) for developing complex interventions from conceptualisation through to implementation of the intervention in community settings aimed at improving health outcomes. The UKMRC guidelines provide a stepwise, phase-based approach to assist researchers with the design and evaluation of complex interventions. Campbell et al. (2007, p. 455) define complex interventions as being “...built up from a number of components, which may act both independently and interdependently”. Furthermore, Craig et al. (2013) provide some clear examples, stating complex interventions comprise of any (or a combination) of the following aspects: 1) a number of interacting components within the experiment and control interventions; 2) multiple and complex behaviours required by those delivering or receiving the intervention; 3) a number of groups or organisational levels targeted by the intervention; 4) multiple outcomes; and 5) a degree of flexibility or tailoring of the interventions is permitted. Based on this, the TOBY app met the criteria to be a complex intervention, thus the UKMRC guidelines were adopted for its evaluation.

The framework consists of four phases that can be utilised either sequentially or iteratively (see Figure 1.1). The four phases can be summarised as follow: 1) the development phase, where the components of the interventions are defined; 2) the feasibility or piloting phase, where the trial and intervention design are defined, including the appropriateness and feasibility of the intervention; 3) the evaluation phase, with the use of an RCT to evaluate effectiveness; and 4) the implementation phase, where the effective long-term implementation of the intervention into practice is completed, including long-term follow-up.





**Figure 1.1 Key elements of the development and evaluation process in the UKMRC Framework**

Prior to the commencement of my PhD research, two papers had been published detailing preliminary investigations related to the development phase, and to a lesser extent, the feasibility and piloting phase of the TOBY app. The work by Venkatesh et al. (2013) clearly articulates the theoretical underpinnings of the TOBY app using ABA principles, while the study by Moore et al. (2015) reported on some aspects of the feasibility of implementing the TOBY app, indicating the TOBY app may make a useful contribution to early intervention for children with ASD by increasing the training hours young children with ASD receive at home with their parents or caregivers. Importantly for my research, the feasibility and piloting by Moore et al. (2015) did not include child-based outcome measures of the skills the TOBY app was targeting. Furthermore, the study had been completed with families who were mostly from Australian city areas, not from regional and remote areas; thus its effectiveness in addressing service access issues remained unknown.

Lastly, in the UKMRC guidelines, Craig et al. (2013) stressed that strict standardisation for each participant in the study might not be appropriate when evaluating an intervention, instead, suggesting interventions may work best when tailored to the local context rather

than maintaining complete standardisation. This point was particularly pertinent in the context of my research. The local context of each of the families with a child with ASD living in regional and remote areas in Australia varies widely, and at least anecdotally the services and supports available to their major city-dwelling counterparts were different.

### **1.6.2 Appropriateness**

The UKMRC guidelines emphasise the importance of assessing the appropriateness of the intervention from the perspectives of the end-user (Campbell et al., 2000; Craig et al., 2008; Hammell, 2001). This is further reinforced in the literature with Nastasi et al. (2000) highlighting the need for intervention developers to assess the appropriateness of the intervention, in addition to its effectiveness. While the UKMRC guidelines highlight the importance of assessing an intervention's appropriateness, the guidelines lack specificity on what aspects of appropriateness should be evaluated. Therefore, an appropriateness framework commonly applied in allied health and therapeutic interventions as described by Evans (2003) was used to evaluate the TOBY app.

Evans (2003) defines appropriateness as being concerned with the psychosocial aspects of the intervention and addresses questions related to the intervention's impact on a person and whether it would be used by the consumer. Appropriateness is an essential step in the development of a complex intervention as, regardless of the intervention's effectiveness, if deemed unacceptable by the end-user, poor adherence and early abandonment of the intervention may occur, thus reducing the overall value of the intervention (Evans, 2003; Solish & Perry, 2008).

Appropriateness, as applied to the context of my research, relates to the perceptions of the parent in relation to the intervention and consists of the following five dimensions: 1) parents perceive the intervention as a positive experience for them and their child; 2)

parents perceive the challenges their child experiences due to their ASD as relevant and important; 3) parents perceive the outcomes of the intervention as beneficial; 4) the components of the intervention are ecologically valid (logistically viable in the parent's and their child's everyday contexts); and 5) strategies developed by using the intervention are continued to be used by the parents once the intervention has ceased (Allan, Wilkes-Gillan, Bundy, Cordier, & Volkert, 2018; Bowen et al., 2009; Cordier et al., 2016; Nastasi et al., 2000; Wilkes-Gillan, Bundy, Cordier, Lincoln, & Hancock, 2015). In relation to my research, the population living in regional and remote WA may have some experiences dissimilar to parents living in major cities in WA, due to lack of health infrastructure to support the implementation of the intervention. To date, no investigations have been conducted evaluating the TOBY app's appropriateness; hence, the investigation into the appropriateness for this population is warranted (Dew et al., 2013; Dew et al., 2012).

While the UKMRC framework and appropriateness aided in guiding the evaluation the TOBY app, there is not enough scope within the two frameworks to provide clear clinical implications and future recommendations to increase the chances of successful implementation and widespread use within the ASD community. Hence, there is a need for an additional theoretical framework to guide practice.

### **1.6.3 Diffusion of Innovation Theory**

When undertaking ICT-based research for the ASD population, and more specifically, how ICT is adopted in interventions for children with ASD, I was surprised by the large amount of efficacy studies in the literature for technology-based interventions for children with ASD. Moreover, I was surprised by the lack of research regarding the implementation and adoption of ICT-based interventions by clinicians and parents of children with ASD in the community. Given the preliminary evidence suggesting that ICT-based interventions have

some utility in ameliorating some of the challenges children with ASD experience and the considerable time and financial investment in developing and evaluating them, it seems a logical next step for researchers to investigate the most effective ways that their users could adopt these interventions. Given the lack of translational research available, combined with the innovative nature of the TOBY app and varying engagement levels by participants as described in the pilot trial by Moore et al. (2015), I needed to incorporate an additional theory to help guide my research, to explain the findings and provide recommendations for future research and clinical practice.

One such theory that helps to understand and explain the phenomenon of innovation adoption is the Diffusion of Innovation theory. It should be noted that while the Diffusion of Innovation theory is applicable to ICT-based interventions, it is also relevant to all innovations, not just those that are technological in nature (Lyytinen & Damsgaard, 2001). The Diffusion of Innovation is a theoretical framework that helps describe how, why, and at what rate innovations are adopted through social systems (Rogers, 2003). The Diffusion of Innovation theory is comprised of four main elements: 1) the innovation; 2) communication channels; 3) time; and 4) a social system. The Diffusion of Innovation theory does have some acknowledged shortfalls, namely being too simplistic and descriptive (Lyytinen & Damsgaard, 2001; Minishi-Majanja & Kiplang'at, 2005). However, when applied to a specific innovation in a defined context, the Diffusion of Innovation theory provides a robust framework to identify the conditions for successful adoption of innovations (Lyytinen & Damsgaard, 2001).

Diffusion is defined as “The process by which an innovation is communicated through certain channels over time among the members of a social system” (Rogers, 2003, p. 10). Moreover, Rogers (2003) emphasises that the social context in which an innovation is

received, strongly influences its adoption. Dingfelder and Mandell (2011), who are one of the few authors to apply the Diffusion of Innovation theory to translating ASD research into practice, reinforced the importance of the social context in the diffusion of evidence-based interventions for children with ASD. Given the unique social context within which my research was undertaken and the “innovativeness” (i.e., affinity to the adoption of new ideas compared to other individuals) of the TOBY app, the Diffusion of Innovation is an appropriate theory to help understand and interpret my findings. While the Diffusion of Innovation theory consists of four broad elements and takes a macro perspective of the spread of an innovation over time, a sub-process of diffusion known as *adoption* is more useful in the context of my research. Adoption refers to an individual’s decision whether to integrate (or reject) a particular innovation in their lives (Straub, 2009). In the Diffusion of Innovation theory, this sub-process of adoption is referred to as the innovation-decision process.

#### **1.6.3.1 Innovation-Decision Process**

The innovation-decision process is one key aspect of the Diffusion of Innovation theory that is useful when trying to understanding how individuals evaluate and adopt an innovation (Rogers, 2003). According to Rogers (2003), the innovation-decision process outlines how, over time, individuals will make choices or complete actions to evaluate an innovation and make a decision on whether or not to incorporate it into their ongoing practice. The innovation-decision process consists of the following five sequential stages: 1) knowledge; 2) persuasion; 3) decision; 4) implementation; and 5) confirmation (see Figure 1.2). In relation to the adoption of the interventions for ASD, the innovation-decision process explains the process by which individuals (therapists, parents) would decide to accept, adopt, or reject the intervention.

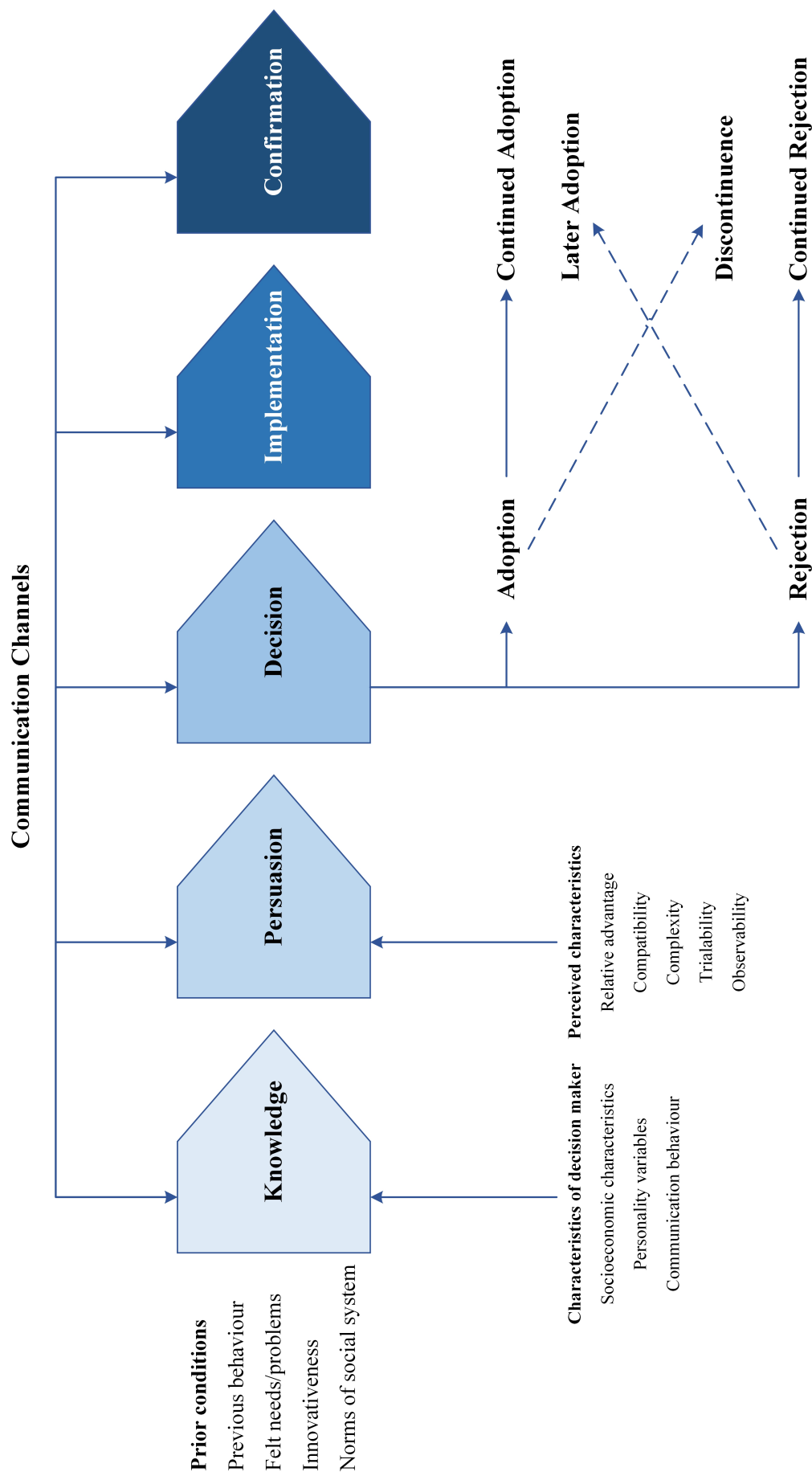


Figure 1.2 Innovation-Decision Process (Rogers, 2003)

The innovation-decision process begins with the knowledge stage. In this stage, the individual becomes aware of a particular innovation and learns more about its functions (awareness-knowledge). For example, in the context of a parent of a child with ASD and a new intervention, this is when the parent becomes aware of a new innovation (intervention) and what it involves. Importantly, prior conditions strongly influence a parent's awareness-knowledge of an intervention. These prior conditions for parents consist of their previous behaviour, their felt need, their affinity for adopting new interventions (their "innovativeness"), and the social norms within their context (Rogers, 2003). Furthermore, the characteristics of individual parents will influence their awareness-knowledge of a new intervention. That is, the socioeconomic, personality and communication behaviour will all impact how each parent becomes aware of an intervention and how they learn more about how it functions.

The next stage is persuasion, whereby the individual develops an attitude (either positive or negative) towards the innovation. Importantly, the characteristics of the innovation are used by individuals to form their attitude. Thus, the characteristics of the innovation are very important in generating an individual's attitude towards an innovation. In the context of a parent of a child with ASD considering the adoption of a new intervention, this is where the parent will determine their attitude towards the intervention. There are five characteristics of an innovation influencing its adoption: 1) relative advantage, the perception that the innovation is better than the one it precedes; 2) compatibility, the perception that the innovation is congruent with the needs, values and experiences of the adopter; 3) complexity, the perceived difficulty to use an innovation; 4) trialability, the innovation's ability to be trialled by the user for a limited period; and 5) observability, the outcomes of adopting the innovation can be substantiated by others (Rogers, 2003).

According to Diffusion of Innovation theory, innovations that are perceived by individuals

as having a greater relative advantage, compatibility, trialability, observability, and less complexity will be adopted more rapidly than other innovations (Rogers, 2002).

The decision stage follows the persuasion stage. The decision stage occurs when an individual undertakes activities that influence their choice on whether to adopt or reject the innovation. This adoption occurs when the parent makes a decision that the intervention is the best course of action available to them and their child. Put simply, will the parent choose to use the new ASD intervention or not? Most people will not adopt an innovation in its entirety, as a mechanism to address the inherent uncertainty associated with adopting something novel. Full adoption is often preceded by partial adoption, and an innovation that can be trialled is often adopted more readily than one that cannot. For interventions for children with ASD, the ability to partially trial the intervention prior to committing fully will help the parents decide if this intervention has a higher relative advantage than other interventions available to them. An example could be that one module of an intervention is offered free-of-charge to families to determine if the child engages with the intervention.

Following the decision stage is the implementation stage. Until the implementation stage, the decision process has primarily been a mental process. However, in the implementation stage, behaviour change is required to put the innovation into practice. Thus, the implementation phase is where problems with actually using the innovation are uncovered. For a parent with a child with ASD starting a new intervention, examples could be where the child does not engage with the activities included in the intervention, or the parent is unable to find the time in their daily life to complete the required activities. A key characteristic of this stage is the existence of uncertainty with the consequences of the innovation. Parents may start asking questions such as “How do I use the intervention?” and “How do I get my child to stay engaged with the intervention?” Importantly for



intervention researchers and developers, there is a high level of information gathering by parents occurring in this stage. Parents need to be able to have their questions answered and, in doing so, will increase the likelihood of adoption of the intervention.

Last is the confirmation stage, whereby the individual seeks out the assurance of their decision or may choose to reverse their decision if they receive conflicting or negative information regarding the intervention. The rejection of an innovation can occur at a later stage, even after the decision to adopt the innovation has been made (represented by the dotted line in Figure 1.2). Rogers (2003) refers to this as discontinuance. Discontinuance can occur because either a better innovation supersedes it (i.e., replacement discontinuance) or an individual elects to reject the innovation after initial adoption due to dissatisfaction with it (i.e., disenchantment discontinuance) (Rogers, 2003). For a parent of a child with ASD who has adopted a new intervention, discontinuance could occur as a new intervention becomes available that they perceive to be superior, or they find that after a period of time they are seeing no improvement in their child's skills so decide to discontinue the intervention. Conversely, individuals who initially reject the decision to adopt an innovation may, in time, elect to adopt the innovation in what is referred to as later adoption (represented by the dotted line in Figure 1.2). Parents may never trial and adopt a new intervention and are therefore deemed to have undertaken continued rejection.

Communication channels are a critical aspect at each stage of the innovation-decision process, providing input and influencing the likelihood of adoption (or rejection) throughout the process. Communication in the Diffusion of Innovation theory relates to how mutual understanding by participants is achieved through the creation and sharing of information relative to the innovation (Rogers, 2003). That is, how the diffusion of the innovation occurs as a result of the information exchange (communication) between one

individual and another or many others. A key aspect of information exchange is the existence of communication channels. Communication channels are the mechanisms through which two individuals can exchange that information. For a parent of a child with ASD, this refers to how they will receive information from the therapists, educators or the intervention developers and how the parent communicates back to these parties.

Communication channels can take many forms, including mass media or individual communication.

Relevant to my research, the Diffusion of Innovation theory and innovation-decision process can assist in explaining why some evidence-based interventions are not utilised in practice for children with ASD, and why interventions with minimal research support are utilised broadly and gain acceptance within the ASD community (Dingfelder & Mandell, 2011).

## **1.7 Overall Aim**

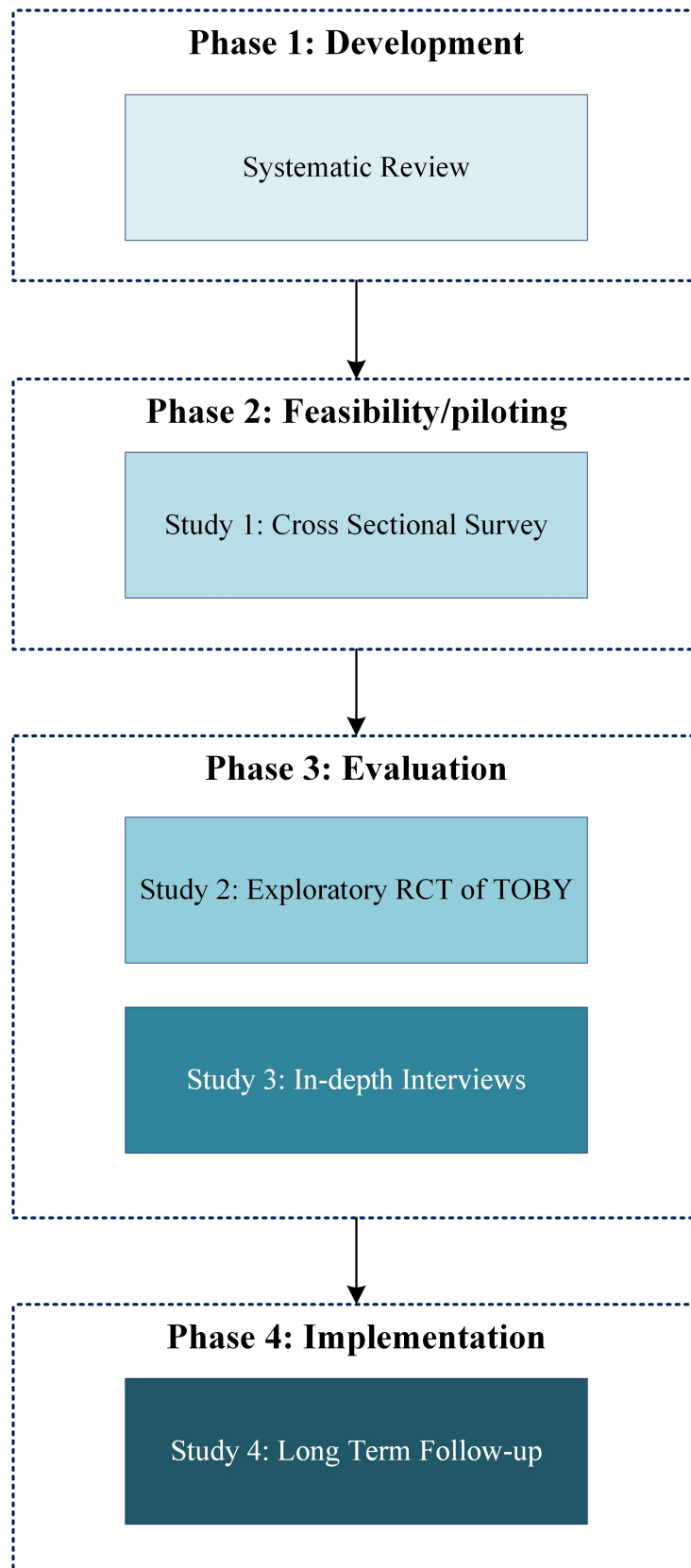
The overall aim of this thesis was to examine the feasibility, effectiveness and appropriateness of an ICT-based intervention, the TOBY app, for children with ASD living in regional and remote WA.

The outcomes of the research will: 1) synthesise the existing research for parent-mediated interventions for families living outside of major cities; 2) compare the coping strategies, stress levels, QoL and daily routines of families in regional and remote WA who have a 2-18 year old child diagnosed with ASD with families from major cities who have a child with diagnosed with ASD; 3) evaluate the effectiveness of the TOBY app as a complementary intervention to existing therapy for children aged 2-6 years with ASD living in regional and remote WA; 4) explore the appropriateness of the intervention in the context of parents who have a child with ASD living in regional and remote WA; and 5)

evaluate the long-term maintenance of skills for the children who participated in the effectiveness trial.

## **1.8 Thesis Structure**

The thesis comprises of two traditional thesis chapters (Introduction chapter, and a Discussion and Conclusion chapter) that bookend the remaining five chapters, which are presented as journal manuscripts. The thesis is structured in four phases of research that is aligned with UKMRC guidelines (see Figure 1.3). Phase One of the research aligns with the development phase of the UKMRC guidelines, consisting of a systematic review (Chapter 2). Phase Two comprises of a cross-sectional survey (Chapter 3) aligning with the feasibility phase of the UKMRC guidelines. Phase Three addresses the evaluation phase of the UKMRC guidelines and comprises an exploratory randomised controlled trial (Chapter 4) and semi-structured interviews with parents who have used the TOBY app within the RCT to examine the appropriateness of the intervention (Chapter 5). Phase Four comprises a long-term follow-up study with participants from the intervention study to measure maintenance of skills learnt (Chapter 6) and is aligned with the final stage of the UKMRC guidelines, the implementation phase. The thesis ends with a Discussion and Conclusion chapter (Chapter 7) that synthesises overall findings. Chapter 7 provides implications and recommendations for the development of ICT-based interventions for children with ASD, identifies areas for future research, and highlights the limitations of this thesis. References are provided for each chapter. Chapters 2, 3, 4 and 5 are either in press or have been published, while Chapter 6 is currently under review.



**Figure 1.3 Thesis Structure**

### **1.8.1 Phase One: Development of the TOBY app**

Chapter 2 is aligned with the development phase of the UKMRC framework. Chapter 2 consists of a systematic review of the literature to provide a rigorous summary of the existing literature. Chapter 2 contributes to theory development of the TOBY app by providing an overview of existing parent-mediated interventions delivered remotely to the families living outside of major city areas interventions and providing recommendations to guide my research with this population. The research objectives of Chapter 2 were to:

1. Systematically review the existing evidence presented by studies on parent-mediated intervention training, delivered remotely for parents having children with ASD and living outside of major cities;
2. Provide an overview of current parent training interventions used with this population; and
3. Provide an overview of the method of delivery of the parent training interventions used with this population.

### **1.8.2 Phase Two: Feasibility of the TOBY app**

Chapter 3 comprises of a study undertaken to investigate the context within which the efficacy study was undertaken. This chapter contributes to the evaluation of the feasibility of the TOBY app. The objectives of this study were to:

1. Explore whether regionality influences stress, coping, QoL and daily routines for parents of a child with ASD (aged 2-18 years) in WA using validated outcome measures; and
2. Compare stress levels and QoL of parents of a child with ASD (aged 2-18 years) in WA with population norms.

### **1.8.3 Phase Three: Evaluation of the TOBY app**

Phase three of the thesis consists of Chapters 4 and 5. Chapter 4 aimed to evaluate the effectiveness of the TOBY app for families living in regional WA through an RCT. The research objective was to conduct an RCT of the TOBY app as an early intervention to augment existing therapies to improve visual motor, imitation, language and social skills in children with ASD, aged between two to six years who reside in a regional area. The study tested three hypotheses:

1. The change in visual motor, imitation, language and social skills of children with ASD who received the TOBY app for three months will be significantly greater than changes in a waitlisted control group;
2. The overall visual motor, imitation, language and social skills of children with ASD who received the TOBY app for three months will improve significantly from baseline; and
3. The overall visual motor, imitation, language and social skills of children with ASD who received the TOBY app for three months will maintain these skills after three months cessation of the intervention.

Chapter 5 aimed to explore the appropriateness of the TOBY app from the perspectives of the parents using a robust qualitative research methodology anchored in a theoretical framework. The research objectives were to:

1. Evaluate the appropriateness of the TOBY app for families of children with ASD living in regional Australia;
2. Provide insight into the barriers and facilitators identified by parents who used the TOBY app living in regional Australia; and

3. Provide insight into ICT-based interventions and better inform the development and use of other ICT-based interventions for clinicians, researchers and developers that use parent-mediated interventions to complement existing therapy services.

#### **1.8.4 Phase Four: Implementation of the TOBY app**

The final phase of the research consists of Chapter 6. The research objective of Chapter 6 was to:

1. Conduct a follow-up study for children with ASD living in regional Australia who used the TOBY app for three months in the RCT effectiveness trial to determine if they maintained or continued to improve their language and social communication skills after at least 12 months post-intervention.

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## Chapter 2      Systematic Review

Chapter 2 details the results of a systematic review investigating existing evidence for parent-mediated intervention training delivered remotely for children with autism spectrum disorder living outside of major city areas. This chapter contains an accepted manuscript of an article published in the Journal of Medical Internet Research which is available online:

<https://doi.10.2196/jmir.6651>

The spelling and wording contained within this chapter are that of the published manuscript.

Note: In this manuscript, the term ‘urban’ was used in place of ‘major city’, and the term ‘rural’ was used in place of ‘regional and remote’.



# Journal Manuscript 1

## **Title**

Parent-mediated intervention training delivered remotely for children with Autism Spectrum Disorder living outside of urban areas: a systematic review.

## **Registration**

The systematic review was registered with PROSPERO register: Registration number CRD42015027300

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## 2.1 Abstract

**Background:** Parent training programs for families living outside of urban areas can be used to improve the social behavior and communication skills in children with autism spectrum disorder (ASD). However, no review has been conducted to investigate these programs.

**Objective:** The aim of this study was to (1) systematically review the existing evidence presented by studies on parent-mediated intervention training, delivered remotely for parents having children with ASD and living outside of urban areas; (2) provide an overview of current parent training interventions used with this population; (3) and provide an overview of the method of delivery of the parent training interventions used with this population.

**Methods:** Guided by the preferred reporting items for systematic reviews and meta-analyses (PRISMA) statement, we conducted a comprehensive review across 5 electronic databases (CINAHL, Embase, ERIC, PsycINFO, and Pubmed) on July 4, 2016, searching for studies investigating parent-mediated intervention training for families living outside of urban centers who have a child diagnosed with ASD. Two independent researchers reviewed the articles for inclusion, and assessment of methodological quality was based on the Kmet appraisal checklist.

**Results:** Seven studies met the eligibility criteria, including 2 prepost cohort studies, 3 multiple baseline studies, and 2 randomized controlled trials (RCTs). Interventions included mostly self-guided websites: with and without therapist assistance (n=6), with training videos, written training manuals, and videoconferencing. Post intervention, studies reported significant improvements ( $p<.05$ ) in parent knowledge (n=4), parent intervention fidelity (n=6), and improvements in children's social behavior and communication skills

(n=3). A high risk of bias existed within all of the studies because of a range of factors including small sample sizes, limited use of standardized outcome measures, and a lack of control groups to negate confounding factors.

**Conclusions:** There is preliminary evidence that parent-mediated intervention training delivered remotely may improve parent knowledge, increase parent intervention fidelity, and improve the social behavior and communication skills for children with ASD. A low number of RCTs, difficulty in defining the locality of the population, and a paucity of standardized measures limit the generalization of the findings to the target population. Future studies should investigate the appropriateness and feasibility of the interventions, include RCTs to control for bias, and utilize standard outcome measures.

## **2.2 Introduction**

Autism spectrum disorders (ASD) are characterized by deficits in social communication and social behavior, including problems interpreting nonverbal gestures, difficulty developing age-appropriate friendships, adherence to rigid routines, and adapting to environmental change (American Psychiatric Association, 2013; Happé & Ronald, 2008). In recent years there has been a marked increase in the prevalence of ASD in children with possible reasons cited including (1) advancement in diagnostic procedures, (2) broadening of the diagnosis criteria, (3) increase awareness of ASD, (4) previous diagnosis, and (5) recognition that ASD is a lifelong condition (Matson & Kozlowski, 2011). The current prevalence rate of ASD ranges from 20 per 10,000 to as high as 110 per 10,000 of the global population (Baio, 2012; Baird et al., 2006; Baron-Cohen et al., 2009; Hill, Zuckerman, & Fombonne, 2015; Kim et al., 2011; Kogan et al., 2009; Matson & Kozlowski, 2011; Williams, MacDermott, Ridley, Glasson, & Wray, 2008).

The increasing prevalence of ASD exerts major demands on early intervention services and education institutions resulting in calls for innovative service delivery models and methods (Ramdoss et al., 2012; Vismara, McCormick, Young, Nadhan, & Monlux, 2013). Limited access to adequate health services and a shortage of adequately trained early intervention health and education professionals are of particular concern in regional and remote areas (Hutton & Caron, 2005; Iacono, Humphreys, Davis, & Chandler, 2004; Smith, Humphreys, & Wilson, 2008; Struber, 2004). Families of children with ASD who live in regional and remote areas often experience several barriers to improving the outcomes for their child (Hoogsteen & Woodgate, 2013b), including (1) increased travel distance to suitably qualified clinicians for effective therapy services, (2) delayed diagnosis due to reduced screening programs, and (3) challenges from the inconsistency of health professionals due to high attrition rates and high workforce transition (Hutton & Caron, 2005; Murphy &

Ruble, 2012; Reese et al., 2015; Smith et al., 2008; Struber, 2004). These challenges highlight the need for innovative and alternative early intervention methods for children with ASD and living outside of urban areas.

Effective early intervention requires skilled health and education professionals and places an increased financial and time burden on families to access services (Buescher, Cidav, Knapp, & Mandell, 2014; Horlin, Falkmer, Parsons, Albrecht, & Falkmer, 2014). As a result, parents or caregivers may be required to play a larger role in the provision of therapeutic services for their children with ASD (Hoogsteen & Woodgate, 2013c). To help overcome these barriers, parents can become active agents in the therapeutic process with the appropriate training and ongoing guidance, thereby delivering these interventions to their children in a more consistent manner (McConachie & Diggle, 2007). This is particularly pertinent for families living outside of urban areas where there is often a lack of access to suitably trained clinicians.

The rise of technological advances in information communication technology (ICT) has paved the way for alternative modes of delivery for health interventions. Evidence suggests that services provided by health professionals using ICT have high efficacy in areas of health, such as delivering behavioral treatment for people with anxiety and depression (Reger & Gahm, 2009; Spek et al., 2007). Moreover, evidence for using telehealth and ICT for children and adolescents with ASD is emerging, with preliminary findings suggesting that it has potential benefits in the diagnosis and delivery of interventions with this population (Boisvert, Lang, Andrianopoulos, & Boscardin, 2010; Gwynette et al., 2017; Hepburn, Blakeley-Smith, Wolff, & Reaven, 2015; McDuffie et al., 2013; Northrup, Lantz, & Hamlin, 2016; Ramdoss et al., 2011; Ramdoss et al., 2012; Terry, 2009).

Systematic literature reviews support the use of parent-mediated interventions in children

with ASD (Diggle & McConachie, 2002; McConachie & Diggle, 2007; Meadan & Daczewitz, 2015), as does the use of telehealth in providing education sessions to parents or caregivers who have a child with ASD (Boisvert et al., 2010; Wainer, 2014). No systematic review has been published on parent-mediated interventions for families having a child with ASD and living outside of urban areas. This is a unique population, and whereas similarities may exist between this group and the general population, these cannot be generalized due to distinctive characteristics and the barriers these families experience due to remoteness (Hoogsteen & Woodgate, 2013c).

Evidence suggests that the characteristics of families having a child with ASD and living outside of urban areas are unique; however, categorizing and comparing populations across countries is challenging because of differing definitions and classifications systems. For example, in Canada, all territories outside of an urban area are considered to be rural. Rural areas include those “...having a population of at least 1000 and a density of 400 or more people per square kilometer...” (Statistics Canada, 2015). Similarly, according to the US Census Bureau, rural areas include all population, housing, and territory not contained within an urban area (United States Census Bureau, 2015). However, the Australian Bureau of Statistics (ABS) uses a 5-category classification based on the Australian standard geographical classification system (ASGC) (Australian Bureau of Statistics, 2011). The categories include major cities, inner regional, outer regional, remote, and very remote based on a number of variables including population size and distance by road to service centers.

The purpose of this systematic review was to review the existing evidence for parent-mediated intervention training delivered remotely for parents having a child with ASD and living outside of urban areas. In doing so, this review will (1) provide an overview of the

studies involving the use of parent-mediated intervention training delivered remotely to parents who have a child with ASD, (2) provide an overview of current parent training programs used with this population, and (3) provide an overview of the method of delivery of parent training interventions used with this population.

## **2.3 Method**

### **2.3.1 Protocol and registration**

The systematic review was registered with PROSPERO (registration number CRD42015027300). The preferred reporting items for systematic reviews and meta-analyses (PRISMA) statement guided the methodology and reporting of this systematic review. The statement provides the structure and transparency considered necessary for reporting systematic reviews in areas of health care.

### **2.3.2 Eligibility criteria**

Participants needed to be parents or caregivers of children diagnosed with ASD. With the recent update to the diagnostic and statistical manual of mental disorders (DSM-5), inclusion criteria were expanded to include participants whose children had a diagnosis of autism, Asperger's syndrome, or pervasive developmental disorder not otherwise specified under criteria of the previous DSM-IV (American Psychiatric Association, 1994). Studies were included if the children with ASD were aged under 18 years. Given the discrepancies in definition and classification of urban-rural location between countries, for the purpose of this review, we included only those studies in which the population resided outside of major cities or urban areas and authors explicitly described participants as having limited access to services.

Articles were included if the intervention involved training the parents or caregivers in intervention skills to improve the social behavior and communications skills for their child

with ASD using telehealth (remote delivery) methods. Face-to-face training, which required parents to travel to a center for training were excluded. Studies were excluded if training was provided solely to therapy professionals or teachers. Telehealth interventions delivered directly and solely by clinicians were excluded from the review, as one study explicitly addressing this issue already exists (Boisvert et al., 2010). Various modes of delivery were accepted for inclusion, including, DVDs, videoconferencing, and Web-based content, as long as the method of delivery enabled remote delivery. Articles of any methodological design that met the eligibility criteria were included, as long as they were published in English in International Scientific Indexing (ISI) listed scientific journals.

### **2.3.3 Information sources**

To identify eligible studies, the authors conducted a comprehensive systematic search across 5 electronic databases on July 4, 2016. Databases searched included (1) Cumulative Index to Nursing and Allied Health Literature (CINAHL), (2) Embase, (3) Education Resources Information Center (ERIC), (4) PsycINFO, and (5) Pubmed.

### **2.3.4 Search strategy**

The categories of search terms used were (1) ASD (autism, autism spectrum disorder, pervasive development disorder not otherwise specified, and Asperger's' syndrome) and (2) residing outside of urban areas (rural health, regional health, remote health, telehealth, telemedicine, and videoconferencing) (see Table 2.1). Search limitations included papers published in English only. A free-text search was completed within the listed databases for literature published from June 16, 2014 to July 4, 2016. The search terms and limitations used for the free-text search are outlined in Table 2.1. Manual searches of the following journals were performed: The Journal of Rural Health (United States), Australian Journal of Rural Health, Rural Educator, Journal of Research in Rural Education, Australian and

International Journal of Rural Education and Rural Special Education Quarterly. Finally, manual searches were conducted of the reference lists of articles that met the eligibility criteria.

### **2.3.5 Study selection**

The first author screened titles and abstracts of the entire pool of articles that met the inclusion criteria and removed duplicates. Following the removal of the duplicates, all abstracts were screened independently by 2 authors using the inclusion or exclusion criteria. Full-text articles were sourced for abstracts that met inclusion criteria, and articles that did not meet the inclusion or exclusion criteria were excluded. Agreement between authors was reached on 8 out of the 9 included articles. The remaining disagreement was resolved through discussion and consensus.

### **2.3.6 Methodological quality**

Methodological quality was assessed using the standard quality assessment criteria as described by Kmet, Lee, and Cook (2004). The Kmet appraisal checklist uses a 3-point ordinal system to assess the methodological quality of research papers. The appraisal checklist creates a systematic, quantitative, and reproducible process to assess the methodological quality of a variety of research designs and make comparisons between them. Two authors independently assessed the included articles using the 14-point checklist. Scores were categorized into quality levels: >80% as strong, 70-80% as good, 50-69% adequate, and <50% as limited. The methodological scores are included in Table 2.2. Disagreements in methodological quality existed between the 2 authors in 2 out of the 9 articles and were resolved through discussion and consensus.



**Table 2.1 Search Terms**

Database and Search terms	Limitations	Number of abstracts
<b>Subject Headings:</b>		
CINAHL: (MH “autistic disorder”) or (MH “child development disorders, pervasive”) or (MH “pervasive developmental disorder—not otherwise specified”), and (MH “rural health centers”) or (MH “hospitals, rural”) or (MH “rural population”) or (MH “rural health services”) or (MH “rural areas”) or (MH “services for Australian rural and remote allied health”) or (MH “rural health”) or (MH “rural health personnel”) or (MH “telehealth”) or (MH “telemedicine”) or (MH “videoconferencing”) or (MH “teleconferencing”)	English language	80
ERIC: SU.EXACT(“Asperger syndrome”) or SU.EXACT(“pervasive developmental disorders”) or SU.EXACT(“autism”), and SU.EXACT(“rural population”) or SU.EXACT(“rural areas”) or SU.EXACT(“rural youth”) or SU.EXACT(“rural environment”) or SU.EXACT(“rural education”) or SU.EXACT(“teleconferencing”) or SU.EXACT(“telecourses”) or SU.EXACT(“videoconferencing”) or SU.EXACT(“telecommunications”)	English language	29
Embase: autism or Asperger syndrome or “pervasive developmental disorder not otherwise specified,” and (rural health care or rural area or urban rural difference or rural population) or (teleconsultation or telediagnosis or telehealth or telemedicine or telemonitoring or teletherapy or videoconferencing or teleconference or health care delivery)	English language	406
PsycINFO: autism or pervasive developmental disorders or Aspergers syndrome, and (exp rural environments or distance education) or (telemedicine or computer mediated communication or telecommunications media)	English language	64
PubMed: (“Autistic disorder” [Mesh] or “child development disorders, pervasive” [Mesh]) and (“rural population” [Mesh] or “rural health services” [Mesh] or “rural health” [Mesh] or “remote consultation” [Mesh] or “telemedicine” [Mesh] or “videoconferencing” [Mesh])	English language	45
CINAHL: Autis* or Asperg* or ASD or (“pervasive,” “developmental,” and “disorder”) or PDD, and (rural* or remote* or regional* or telehealth or tele-health or telemedicine or tele-medicine or telerehab* or tele-rehab* or telediagnos* or tele-diagnos* or teletreat* or tele-treat or teletherap* or tele-therap* or telemonitoring or tele-monitoring or teleintervention or tele-intervention or teletreatment or tele-treatment or telepractice or tele-practice or videoconference* or video-conferenc* or teleconference* or tele-conference* or webbased OR web-based or internet-based or [“technology” and “mediated”] or technology-mediated)	English Language  Published date: 20140601-20160704	64
<b>Free-text search words</b>		
ERIC: As per CINAHL free text	As per CINAHL free text	45
Embase: As per CINAHL free text	As per CINAHL free text	487
PsycINFO: As per CINAHL free text	As per CINAHL free text	131
PubMed: As per CINAHL free text	As per CINAHL free text	446

### **2.3.7 Data collection**

Data were extracted using comprehensive data extraction forms and grouped under the following headings: (1) aims or objectives, (2) study design, (3) level of evidence, (4) participant characteristics (including geographical location and proximity to services), (5) intervention characteristics, (6) outcome measures, (7) discussion, (8) limitations, and (9) implications for future practice. Data extraction was undertaken by the first author. Data extracted was checked by a second author for accuracy. Only minor discrepancies occurred, and these were resolved through consensus. The level of evidence was determined using the hierarchy of evidence as outlined in the National Health and Medical Research Council (NHMRC) guidelines (National Health and Medical Research Council, 1999). Additionally, details of the intervention, dosage, method of delivery, and skills or aims being delivered by the researchers were extracted and summarized. Few studies included in the review had large sample sizes, and the lack of control or comparison groups prevented a meta-analysis.

### **2.3.8 Data items, risk of bias and synthesis of results**

Participant characteristics were extracted and are represented in Table 2.2. Kmet ratings were used to assess the risk of bias of at an individual study level (Kmet et al., 2004). The extrapolated data from this process are represented in Table 2.3. Characteristics of the extracted data included (1) aims and objectives, (2) study design, (3) level of evidence, (4) intervention characteristics, (5) outcome measures, (6) results, and (7) methodological quality. Significance of data and calculated effect sizes of the interventions were extracted for synthesis. Effect sizes not reported as Cohen  $d$  were converted for uniformity as appropriate. The magnitude for Cohen  $d$  effect sizes was interpreted as small  $\geq 0.20$ , medium  $\geq 0.50$ , or large  $\geq 0.80$  (Cohen, 1992). None of the researchers authored any of the included published studies; hence, no bias in study selection was introduced in conducting

the systematic review.

## 2.4 Results

### 2.4.1 Study selection

The PRISMA diagram is presented in Figure 2.1. Database searches yielded 1797 articles. Four additional articles were identified through manual searches of the included studies' reference lists. From the 2001 articles, 583 duplicates were removed, leaving a total of 1218 abstracts for screening. Following screening of the abstracts, 1202 articles were excluded. The remaining 16 were retrieved for full-text review, and an additional 7 articles were excluded from the study, as participants in four studies were not described as living outside of major cities or urban areas.

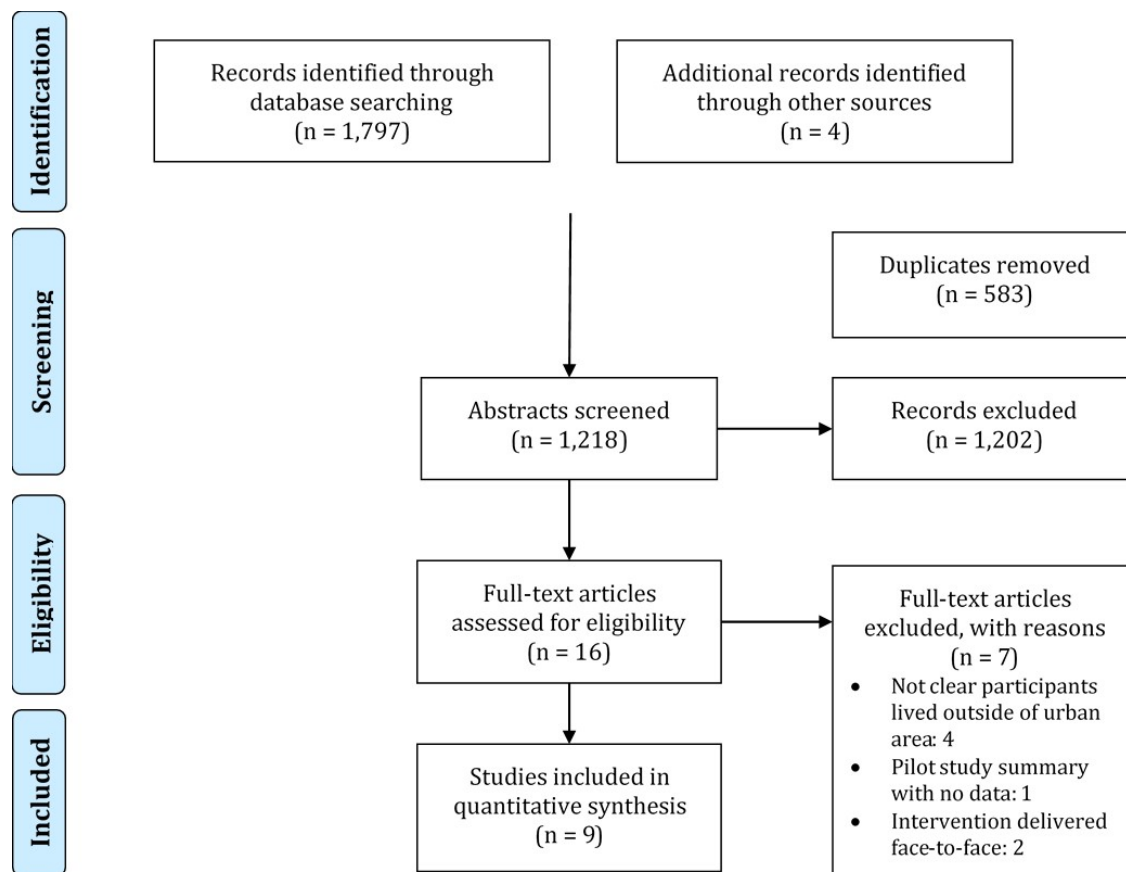


Figure 2.1 PRISMA diagram

One study was a summary of a pilot project with no results included, and the 2 remaining studies provided the training to parents in a face-to-face medium. A total of 9 articles met the review eligibility criteria. The articles by Ingersoll and Berger (2015), Ingersoll, Wainer, Berger, Pickard, and Bonter (2016), and Pickard, Wainer, Bailey, and Ingersoll (2016) were based on the outcomes from one study, and so they were combined for reporting and discussion throughout this paper.

#### **2.4.2 Study design**

The 7 studies included 1 quasi-experimental design by St. Peter et al. (2014), 1 nonconcurrent multiple-baseline designs by Wacker et al. (2013), 2 single-subject multiple-baseline design by Vismara et al. (2013); Vismara, Young, and Rogers (2012), 1 RCT each by Ingersoll et al. (2016) and Pickard et al. (2016), and 2 prepost-test design studies by Hamad, Serna, Morrison, and Fleming (2010) and Heitzman-Powell, Buzhardt, Rusinko, and Miller (2014). A lack of control groups in 5 of the 7 studies precluded the ability to conduct a meta-analysis of the results. An overview of the included papers is provided in Figure 2.2.

Citation	Study Design			Participants		Delivery Method		Aims		OM <sup>a</sup>	ES <sup>b</sup>	Interventions – research created or established	
	Pre-Post	Multiple-base design	RCT <sup>c</sup> / QE <sup>d</sup>	Parents	Child	Online delivered	Face-to-face	Increase parental ASD <sup>e</sup> knowledge	Parent-mediated intervention	Outcome measures created by researchers	Reported Effect Sizes	Researcher Created	Established
Hamad et al. [47]	●			●		●		●	●	●	●	●	
Heitzman-Powell et al. [48]	●			●		●		●	●	●		●	
Ingersoll and Berger [44]			●	●		●		●	●	●		●	
Ingersoll et al. [45]			●	●	●	●		●	●	●	●	●	
Pickard et al. [46]			●	●		●		●	●	●		●	
St. Peter et al. [41]			□ <sup>f</sup>	●					●	●		●	●
Vismara et al. [43]		x <sup>g</sup>		●	●	●		●	●	●	●	●	●
Vismara et al. [12]		x		●	●	●		●	●	●	●	●	●
Wacker et al. [42]		† <sup>h</sup>		●	●	●		●	●	●		●	●

<sup>a</sup>OM: outcome measures.

<sup>b</sup>ES: effect sizes.

<sup>c</sup>RCT: randomized controlled trial.

<sup>d</sup>QE: quasi-experimental.

<sup>e</sup>ASD: autism spectrum disorder.

<sup>f</sup>□= quasi-experimental.

<sup>g</sup>x= single subject design.

<sup>h</sup>†= Nonconcurrent design.

**Figure 2.2 Study schema**

**Table 2.2 Participant characteristics**

Study	No. of participants	Geographical location	Demographics: parent	Demographics: child
Hamad et al. (2010)	51	“Geographically disparate” in the United States	Gender: male n=4, female n=47 Age: not specified Education level: high school n=6, associate degrees n=0, bachelor degrees n=20, master degrees n=12, other n=3 Note: subgroup demographic breakdown not provided	Gender: not specified Age: not specified
Heitzman-Powell et al. (2014)	7	Remote areas in the United States	Gender: not specified Age: mean age 37.3 (range=32-47) Education level: ranged from graduate degree to high school diploma. Breakdown not specified.	Gender: not specified Age: not specified
Ingersoll and Berger (2015)	27	70% (19/27) of participants resided in “rural or medically underserved areas”	Gender: male n= 1, female n=26 Age: not specified Education: college degree or higher=16, education levels of remaining participants not specified	Gender: male n=19, female n=8 Age: mean chronological age 3 years, 6 months.
Ingersoll et al. (2016)				
Pickard et al. (2016)				
St. Peter et al. (2014)	32	Rural Appalachian counties in West Virginia, Kentucky, Maryland, Virginia, or Pennsylvania, United States	Gender: male n=11, female n=21 Age: mean age of parents=35.87 years (range, 24-69) Education level: 54.15% had received a college degree. Remaining participant education levels not specified.	Not specified
Vismara et al. (2012)	8	“Very little access to early intervention services” in California, North Carolina,	Gender: male n=1, female n=7 Age: not specified Education: not specified	Gender: male n=7, female n=1 Age: mean chronological age 2 years, 4 months (standard

Study	No. of participants	Geographical location	Demographics: parent	Demographics: child
		Arkansas, Texas, and Pennsylvania, United States.		deviation=7.6 months, range 16-38 months)
Vismara et al. (2013)	8	“Minimally available intervention services in their community” in the United States and Canada	Gender: male n=1, female n=7 Age: not specified Education level: college n=4, post-college n=4	Gender: not specified Age: 1 year n=4, 2 years n=2, 3 years n=1
Wacker et al. (2013)	17	Regional Iowa, United States	Gender: male n=2, female n=16 Age: mean age 33 years Education level: “most” had some level of postsecondary education. Breakdown not specified.	Gender: male n=16, female n=1 Age: 2 years n=3, 3 years n=4, 4 years n=3, 5 years n=5, 6 years n=2 (range 29-80 months)

**Table 2.3 Study Characteristics**

Citation and methodology	Aim or objectives	Outcome measures	Results	Methodological quality
Hamad et al. (2010)  Pre- or post-test	Investigate the feasibility of an Internet-based “asynchronous” small-scale three module-Web-based learning course presented in a distance-learning medium.	Parent outcome measures: 25-item Web-based knowledge acquisition measure (test) administered prepost intervention.          Child outcome measures: not specified	Internet-based training curriculum could be effective in training parents about methods and procedures related to behavioral interventions.   Pre-test scores: mean=68.8, SD=15.6, Post-test scores: mean=82.9, SD=4.9. Large effect size (Cohen $d=1.21$ )   Paired $t$ -tests: mean prepost-test scores statistically significant improvement ( $p<.001$ ) for all participants combined (n=51).	Kmet rating: strong (82%)  NHMRC <sup>a</sup> level of evidence: level IV
Heitzman-Powell et al. (2014)  Pre- or post-test	Evaluate the modified OASIS training intervention for use with parents from a distance.	Parent outcome measures: parent skill assessment in ABA <sup>b</sup> implementation      Parent knowledge assessment (Web-based) on ASD <sup>c</sup> and ABA principles and procedures  Parent satisfaction with training	Implementations of ABA skills (41.23% mean increase)      Knowledge assessments (39.15% mean increase)   High levels of importance and significance of Web-based tutorials (mean scale 1-5:4.62	Kmet rating: good (77%)  NHMRC level of evidence: level IV



Citation and methodology	Aim or objectives	Outcome measures	Results	Methodological quality
			and 4.71 respectively). High levels of importance and significance of telemedicine coaching sessions (mean scale 1-5:4.62 and 4.8 respectively)	
		Cost savings (driving miles)	Mean travel savings per family was 2,263 driving miles using telemedicine if compared with face-to-face coaching.	
			Note: Prepost comparison with no statistical analysis for significance.	
		Child outcome measures not specified		
Ingersoll and Berger (2015)	Compare parent engagement and effectiveness in self-directed and therapist-assisted versions of a novel telehealth-based parent-mediated intervention for young children with ASD	Parent outcome measures: ImPACT knowledge quiz: 20-item multiple choice quiz taken pre-post intervention	Intervention completion was a significant predictor of post intervention knowledge ( $p=.01$ ) in both groups.	Kmet rating: strong (85%)  NHMRC level of evidence: level II
Ingersoll et al. (2016)				
Pickard et al. (2016)				
RCT <sup>d</sup>		Videotape parent-child interaction for intervention fidelity using the ImPACT intervention fidelity checklist	Intervention completion ( $p=.3$ ) and group assignment ( $p=.45$ ) made significant independent contributions to treatment fidelity. Post intervention fidelity for both groups was significant ( $p=.004$ )	

Citation and methodology	Aim or objectives	Outcome measures	Results	Methodological quality
			Statistically significant improvement pre-post in parent intervention fidelity in both groups ( $p<.01$ , Large effect size: Cohen $d=3.21$ ) as well as between groups post intervention ( $p<.01$ , Large effect size: Cohen $d=0.3$ ). At follow-up statistically significant ( $p<.001$ , Large effect size: Cohen $d=2.92$ ) pre-post in both groups but not between groups.	
		Parent sense of competence scale	Statistically significant improvement ( $p<.01$ , Large effect size: Cohen $d=1.34$ ) prepost intervention in self-efficacy in both groups but not between groups.	
		Family impact questionnaire	Statistically significant improvement ( $p<.05$ , Large effect size: Cohen $d=1.03$ ) prepost in parent stress in both groups but not between groups post intervention. Statistically significant improvement ( $p<.05$ , Large effect size: Cohen $d=1.47$ ) prepost in positive perception of the child in both groups as well as between groups post intervention ( $p<.05$ , Large effect size: Cohen $d=1.16$ ).	
		Parent engagement using website analytics	Therapist-assisted group statistical significantly performed better on parent engagement (number of logins and duration	

Citation and methodology	Aim or objectives	Outcome measures	Results	Methodological quality
			on site) and intervention completion when compared with self-directed groups ( $p<.001$ and $p<.05$ respectively)	
		Intervention evaluation survey using 7-point Likert scale measuring treatment appropriateness, website usability, and overall intervention satisfaction.	Participants rated intervention as highly acceptable (mean=6.07, SD=0.79), the website as highly usable (mean=6.36, SD=0.57). Overall satisfaction of intervention was high (mean=6.56, SD=0.71). No statistically significant difference in treatment appropriateness, website usability, and overall intervention satisfaction between groups.	
		49-item 7-point Likert scale quantitative survey administered post intervention examining appropriateness perceived child social communication gains, burden of the intervention on the family, and frequency of intervention use.	Overall, parent rated intervention favorably with mean scores:  Intervention appropriateness 6.59 (SD 0.58), perceived child social communication gains 5.41 (SD 1.24), burden of the intervention on the family 5.72 (SD 1.23), frequency of intervention use 6.36 (SD 0.57)  Statistically significant differences between groups (TA <sup>e</sup> vs SD <sup>f</sup> ) for intervention appropriateness ( $p=.03$ , Large effect size: Cohen $d=0.94$ ) and child social communication gains ( $p=.05$ , Large effect size: Cohen $d=0.84$ ). No difference in the burden of intervention on the	

Citation and methodology	Aim or objectives	Outcome measures	Results	Methodological quality
			family and frequency of intervention use domains.	
		Qualitative interviews—semistructured investigated overall perception of intervention and content, perception of feasibility of intervention, experience of support during intervention, and intervention referral preferences.	Qualitative themes: Positive perception of the appropriateness of intervention. The intervention was easy to learn initially but became more challenging as they progressed.  The support of a coach would be essential in the later, more complex sections of the intervention.	
			Parents felt more empowered and better able to interact with their child. Perceptions of barriers included time restrictions and technology failure. Parents suggested the intervention should be made available at the time of ASD diagnosis as it may help empower parents at a stressful time.	
		Child outcome measures: language targets	Statistically significant ( $p<.05$ , Large effect size: Cohen $d=2.26$ ) prepost improvements in language targets in both groups but not between groups post intervention.	
		MacArthur communicative development inventories: words and gestures	Statistically significant ( $p<.01$ , Large effect size: Cohen $d=1.74$ ) pre-post improvements in language skills in both groups but not between groups post intervention.	

Citation and methodology	Aim or objectives	Outcome measures	Results	Methodological quality
		Vineland adaptive behavior scales, 2 <sup>nd</sup> edition	Statistically significant ( $p<.05$ , Large effect size: Cohen $d=1.00$ ) prepost improvements in the communication domain in both groups but not between groups post intervention. No statistically significant differences pre-post in the social domains in both groups, however, a statistical difference was observed between groups post intervention ( $p<.05$ , Large effect size: Cohen $d=0.91$ )	
St. Peter et al. (2014)	Compare parental adherence during written or asynchronous video teleconsultation designed to teach parents of children with ASD to implement discrete trial instruction.	Parent outcome measures: Parental adherence between the written (control) and video (experimental) groups	Adherence in the video group was significantly higher ( $p<.001$ ) compared with written instructions.	Kmet rating: good (71%)
Quasi-randomized		Child outcome measures: not specified		NHMRC level of evidence: level III-1
Vismara et al. (2012)	To assess if a 12-week videoconferencing and DVD learning module (P-ESDM <sup>®</sup> ) could improve parents' acquisition of teaching procedures and result in changes in the child's social communicative behavior [55].	Parent outcome measures: Eight item, 5-point response scale evaluating parental satisfaction (feasibility and appropriateness) with the support and ease of the intervention	All parents reported satisfaction with support and ease of the telehealth learning intervention. Six parents identified DVD's as more useful teaching aids compared to handouts. All parents agreed they would recommend an approach to other parents of children with ASD with limited access to community services.	Kmet rating: good (77%)
Single-subject, multiple-baseline design		P-ESDM fidelity tool—5-point Likert rating tool of 13 parent behavior that	Significant increases over time from baseline to follow-up ( $p<.001$ )	NHMRC level of evidence: level IV

Citation and methodology	Aim or objectives	Outcome measures	Results	Methodological quality
		define the child-centred, responsive interactive style used in P-ESDM		
		MBRS <sup>h</sup> —A 5-point Likert rating scale measuring the parent's style of interacting to or relating to their child.	Significant increases in parental behavior rating from baseline to follow-up in responsivity ( $p<.001$ ), affect ( $p<.001$ ), and achievement orientated behavior ( $p<.001$ )	
		Child outcome measures: child social communication behavior—10-min videos transcribed and scored for the production of spontaneous and promoted functional verbal utterances and approximations and imitative play actions on objects and gestures.	Significant overall increases from baseline to follow-up in spontaneous functional verbal utterances ( $p<.001$ ), prompted words over time ( $p<.001$ ), and spontaneous imitations ( $p<.001$ )	
		CBRS <sup>i</sup> [53]—measures engagement and interest in activity as well as joint attention, creativity, and affect demonstrated toward the parent.	Significant increase from baseline to follow-up in child attention ( $p<.001$ ) and child initiation ( $p<.001$ ).	
		MacArthur communicative development inventories: words and gestures	Significant increases from baseline to follow-up with vocabulary production ( $p<.001$ ) and vocabulary comprehension ( $p<.001$ ).	

Citation and methodology	Aim or objectives	Outcome measures	Results	Methodological quality
		Vineland adaptive behavior scales, 2 <sup>nd</sup> edition	Significant increase from baseline to follow-up on the adaptive behavior composite ( $p<.05$ ).	
Vismara et al. (2013)	Pilot study of a 12-week telehealth on the Web (videoconferencing and self-guided website) intervention (P-ESDM) and 3-month follow-up to assess: (1) parents' perception of the intervention as a useful learning platform, (2) parents' intervention skills and engagement style improvement, (3) website utility to support the intervention, and (4) improvements in the children's verbal language and joint attention.	Parent outcome measures: Eight item, 5-point response scale evaluating parental satisfaction with the support and ease of the telehealth learning intervention	All parents reported satisfaction with support and ease of the telehealth learning intervention.	Kmet rating: good (77%)
Single-subject, multiple-baseline design		P-ESDM fidelity tool—5-point Likert rating tool of 13 parent behavior that define the child-centred, responsive interactive style used in P-ESDM	Improvement in parent intervention fidelity. Baseline: 0/8 parents meeting criteria for fidelity in tool. Group mean 2.93 (SD 0.6), post intervention: 6/8 parent meeting criteria for fidelity in tool. Group mean 3.69 (SD.51), follow-up: 7/8 parents achieved at least one fidelity score. Group mean 4.15 (SD 0.51)	NHMRC level of evidence: level IV
		Website use	Average number of logins 30 (SD 18, range 9-60); Average viewing time per day 18 min	
		MBRS [54]—A 5-point Likert rating scale measuring the parent's style of interacting to or relating to their child.	Improvement in parent engagement style. Baseline: low-moderate with MBRS total score mean=2.91, SD=0.68, post intervention: mean=3.50, SD=0.44, follow-up (3 months): moderate to high range with MBRS total score mean=3.87, SD=0.42	
		Child outcome measures:	Increase in the range of vocalizations at all time points	

Citation and methodology	Aim or objectives	Outcome measures	Results	Methodological quality
		behavior scoring of videotaped probes—functional verbal utterances and nonverbal joint attention initiations without gestures	Baseline: mean=2.97, SD=1.93, post interventions: mean=3.60, SD=2.51, follow-up: mean=4.14, SD=2.04  Joint attention initiations remained constant between baseline (mean=1.67, SD=1.07) to post intervention (mean=1.67, SD=1.21) but increased at follow-up (mean=2.16, SD=1.34)	
		MacArthur communicative development inventories: words and gestures	Improvements in VP <sup>j</sup> and comprehension, Baseline: VP mean=111.87, SD=156.03, comprehension mean=224.37, SD=133.25, post intervention: VP mean=163.88, SD=156.03, comprehension mean=284.88, SD=141.53, follow-up: VP mean=213.88, SD=155.08, comprehension mean=314.88, SD= 94.16	
Wacker et al. (2013)	Conduct functional communication training using coaching from trained behavior analysts to parents via telehealth and compare it with completing the same training in-vivo within families' homes.	Parent outcome measures: Parent overall appropriateness—7-point Likert scale	Parents rated training as acceptable (mean=6.47. Comparable with in-vivo training (mean=6.18)	Kmet rating: good (73%)
Nonconcurrent multiple baseline design		Costs: mileage and consultant costs	Costs through telehealth were considerably lower than for in-home behavior therapy	NHMRC level of evidence: level IV
		Child outcome measures: Interobserver agreement on child-targeted problem	Reduction in child-targeted problem behavior when parents coached via telehealth (mean reduction=93.5%). Comparable	



Citation and methodology	Aim or objectives	Outcome measures	Results	Methodological quality
		behavior using interval-by-interval comparisons.	with in-vivo training (mean reduction=94.1%).	

<sup>a</sup>NHMRC: National Health and Medical Research Council. Designation of levels of evidence: I—Evidence obtained from a systematic review of all relevant randomized controlled trials, II— evidence obtained from at least one properly designed randomized controlled trial, III-1—evidence obtained from well-designed pseudo-randomized controlled trials (alternate allocation or some other method), III-2—evidence obtained from comparative studies with concurrent controls and allocation not randomized (cohort studies), case-control studies, or interrupted times series with a control group, III-3—evidence obtained from comparative studies with historical control, two or more single-arm studies, or interrupted time series without a parallel control group, IV—evidence obtained from case series, either post-test or pre-test and post-test.

<sup>b</sup>ABA: applied behavior analysis.

<sup>c</sup>ASD: autism spectrum disorder.

<sup>d</sup>RCT: randomized controlled trials.

<sup>e</sup>TA: therapist-assisted group.

<sup>f</sup>SD: self-directed group.

<sup>g</sup>P-ESDM: parent model—early start Denver model.

<sup>h</sup>MBRS: maternal behavior rating scale.

<sup>i</sup>CBRS: child behavior rating scale.

<sup>j</sup>VP: vocabulary production.

### 2.4.3 Level of evidence

The overall level of the evidence for the studies included in the systematic review was low. The studies by Hamad et al. (2010), Heitzman-Powell et al. (2014), Vismara et al. (2013); Vismara et al. (2012), and Wacker et al. (2013) demonstrated level IV evidence. The study by St. Peter et al. (2014) demonstrated level III-1 evidence and the study by Ingersoll and Berger (2015), Ingersoll et al. (2016), and Pickard et al. (2016) was classified as level II evidence as per the NHMRC level of evidence guidelines (See Table 2.3) (National Health and Medical Research Council, 1999). The low level of evidence may indicate that it is difficult to conduct research with this population due to travelling distance to research centers and reachability through recruitment strategies, thus making robust study designs more challenging.

### 2.4.4 Study participants

For the purposes of this review, study participants were families having a child with ASD, living outside of urban areas, and having limited access to services as reported by the

authors. The inherent difficulty of defining regional and remote localities between different countries made delineating study participants based on geography challenging. None of the studies provided quantitative detail about the participants' proximity and access to services so the interpretation of the findings in relation to this information was impossible. Studies included a total of 197 parents aged between 24 and 69 years involved across the 7 studies.

#### **2.4.5 Outcomes**

The aim of all of the studies was to improve social behavior and communication skills of children with ASD through increasing the knowledge of parents and caregivers by training them in intervention skills (parent-mediated). Outcome measures varied across all of the studies. All 7 studies used measures created by the researchers. Calculated effect sizes were only possible based on the published information in 3 studies included in the review and are reported in Table 2.3.

Parental satisfaction and perceptions of appropriateness of the intervention were measured by Vismara et al. (2013); Vismara et al. (2012), Heitzman-Powell et al. (2014), and the study by Ingersoll et al. (2016) and Pickard et al. (2016). All reported that parents were satisfied with the training they received. When comparing a therapist-assisted and self-guided website versus a self-guided website only, large effect sizes were recorded in parents' perception of the appropriateness of the intervention and child social communication gains (Cohen  $d=0.94$  and  $0.84$  respectively) in the study by Ingersoll and Berger (2015), Ingersoll et al. (2016), and Pickard et al. (2016).

Parents' self-efficacy was evaluated in the study by Ingersoll and Berger (2015) and Pickard et al. (2016). The authors stated that whereas there was a statistically significant ( $P<.01$ ) improvement and a large effect size (Cohen  $d=1.34$ ) preintervention to postintervention for both groups, there was no difference between groups. Parents' stress

levels were not measured prepost interventions in any of the studies.

Knowledge acquisition by parents was measured by Hamad et al. (2010), Heitzman-Powell et al. (2014), and in the study by Ingersoll and Berger (2015), Ingersoll et al. (2016), and Pickard et al. (2016) using quizzes covering the content in the intervention; all studies reported significant increases in knowledge post intervention. Parents' skills in implementing the acquired therapy techniques were investigated by Heitzman-Powell et al. (2014), St. Peter et al. (2014), Vismara et al. (2013); Vismara et al. (2012), Wacker et al. (2013), and in the study by Ingersoll and Berger (2015), Ingersoll et al. (2016), and Pickard et al. (2016). All of the studies reported statistically significant improvements in parents' skills in administering skills learnt through the interventions. These findings present evidence that parents who received the appropriate training could gain skills in the delivery of interventions, thus improving the skills in social communication and behavior of their children with ASD.

Vismara et al. (2013), Vismara et al. (2012), Ingersoll and Berger (2015), Ingersoll et al. (2016), and the study by Pickard et al. (2016) all utilized the MacArthur communicative developmental inventories to measure the child's abilities in vocabulary production and comprehension. In all 3 studies, statistically significant improvements were reported in the children's vocabulary production and comprehension from baseline to follow-up. Again, this provides preliminary evidence that parents who live in geographically isolated areas are able to learn skills in the provision of therapy and implement it appropriately to help improve the communication skills of their children with ASD.

Improvements in social behavior were measured in 2 studies using the Vineland adaptive behavior scales (2nd edition) (Mahoney & Wheeden, 1998) with Ingersoll and Berger (2015), Ingersoll et al. (2016), and Pickard et al. (2016) reporting no significant difference

prepost intervention and Vismara et al. (2012) reporting a significant difference in the social domains. Video-recorded interactions of the children with their parents were used in the studies conducted by Vismara et al. (2013); Vismara et al. (2012) and Wacker et al. (2013). All reported statistically significant improvements prepost intervention in joint attention and affect toward the parents with Wacker et al. (2013) reporting a reduction in child problem behavior.

In summary, it appears that interventions targeting parents' knowledge and including fidelity checks have statistically significant improvements with large effect sizes when reported. Additionally, large to small effect sizes were reported in the child's improvement in social behavior and communication skills when reported within the studies.

#### **2.4.6 Interventions**

All interventions were developed with consideration of the geographical isolation of participants, with the aim to ease administration of the intervention and increase feasibility of delivery. Parent training interventions investigated in the included articles are summarized in Table 2.4.

**Table 2.4 Intervention characteristics**

Study	Intervention description and dosage	Method of delivery to parent	Skills or aims of intervention
Hamad et al. (2010)	Web-based training intervention in behavioral interventions  Dosage: intervention approximately 4-8 h within a 3-week period  Three modules	On the Web using Blackboard Vista 4 platform  Included: short Web-based lectures, practical exercises, video demonstrations of procedures, study questions, and frequent short Web-based quizzes.	<ul style="list-style-type: none"> <li>• Positive reinforcement: selection and use of reinforcement.</li> <li>• Relationship building: parent and teaching cooperation.</li> <li>• Prompting and prompt fading.</li> </ul>
Heitzman-Powell et al. (2014)	OASIS training intervention Research-to-practice Applied behavior analysis outreach training model  Dosage: Eight modules; timeframe not specified	Training program combines Web-based instructional modules and participation in distance coaching sessions.	<ul style="list-style-type: none"> <li>• Introduction to ASD<sup>a</sup> and behavioral treatment;</li> <li>• Basic ABA<sup>b</sup> principles and procedures</li> <li>• Use ABA procedures to teach new skills</li> <li>• Use ABA procedure to reduce challenging behavior</li> <li>• Generalize skills to other settings</li> <li>• Collection and analysis of data for data-based intervention decision-making</li> <li>• Working with treatment teams and other providers</li> </ul>
Ingersoll and Berger (2015)	Project ImPACT on the Web—Website-based training for a naturalistic, developmental-behavioral, parent-mediated intervention for children with ASD	Access to training material was on the Web via personal computer. Included: narrated slideshow with embedded video examples of techniques, written description of lessons, exercises, homework, and reflection questions	<ul style="list-style-type: none"> <li>• Promote child social communication within the context of play and daily routines</li> </ul>
Ingersoll et al. (2016)	ASD  Dosage: Self-directed—Encourage to complete 1 lesson		

Study	Intervention description and dosage	Method of delivery to parent	Skills or aims of intervention
Pickard et al. (2016)	per week, approximately 80 min for 12 weeks.  Therapists assisted—dosage same as self-directed group plus 2 30-min remote coaching sessions per week by trained therapist.	Training program for the therapist-assisted group was administered by trained therapists using videoconferencing software.	
St. Peter et al. (2014)	Implementation discrete-trial instructions using a video training materials  Dosage: video training was 37 min in duration  Written training was a 30-page manual	Written training materials (control) or video training materials (experimental) containing similar content.	<ul style="list-style-type: none"> <li>• Increase adherence to discrete-trial instruction procedures.</li> </ul>
Vismara et al. (2012)	Parent early start Denver model (P-EDSM) training  Dosage: Once-per-week, 1 h parent training sessions for 12 weeks	Telehealth delivery using live, 2-way conferencing with a qualified therapist and the provision of a DVD including all intervention materials with the addition of video recorded examples of the therapist demonstrating skills.	<ul style="list-style-type: none"> <li>• Increasing child's attention and motivation</li> <li>• Using sensory social routines</li> <li>• Promoting dyadic engagements and joint activity routines</li> <li>• Enhancing nonverbal communication</li> <li>• Building imitation skills</li> <li>• Facilitating joint attention</li> <li>• Promoting sequence relations</li> <li>• Employing promoting, shaping, and fading techniques</li> <li>• Conducting functional assessments of behavior to develop new interventions.</li> </ul>

Study	Intervention description and dosage	Method of delivery to parent	Skills or aims of intervention
Vismara et al. (2013)	<p>Parent early start Denver model (P-EDSM) training</p> <p>Dosage: Once-per-week, 1.5 h parent training sessions for 12 weeks</p>	Telehealth delivery using live, 2-way conferencing with a qualified therapist and a self-guided website.	<ul style="list-style-type: none"> <li>• Increasing child's attention and motivation</li> <li>• Using sensory social routines</li> <li>• Promoting dyadic engagements and joint activity routines</li> <li>• Enhancing nonverbal communication</li> <li>• Building imitation skills</li> <li>• Facilitating joint attention</li> <li>• Promoting sequence relations</li> <li>• Employing promoting, shaping, and fading techniques</li> <li>• Conducting functional assessments of behavior to develop new interventions.</li> </ul>
Wacker et al. (2013)	<p>Functional communication training coaching for parents</p> <p>Dosage: Weekly 60 min sessions until completion of treatment,</p>	Telehealth using PC and video-monitors from behavior consultants	<ul style="list-style-type: none"> <li>• Child taught to comply with task request and then to mand for a break to play</li> <li>• Child requesting toys after having to wait for increasing period of time</li> <li>• Request attention when adult attention was removed.</li> </ul>

All interventions were developed by the researchers and varied in dosage and method of delivery. This variation makes synthesis of the research challenging and limits the generalizability of these methods of intervention to the targeted population. Dosage for the interventions ranged from an intensive format of 5 hours per day for 5 days, to once-a-week over a number of weeks. The most common dosage was once-a-week sessions, with sessions lasting 1-2 hours; however, timeframes ranged from 6-12 weeks (Ingersoll & Berger, 2015; Ingersoll et al., 2016; Pickard et al., 2016; Vismara et al., 2013).

Additionally, the studies by Heitzman-Powell et al. (2014) and Wacker et al. (2013) did not have finite periods, and the duration of intervention continued until all training modules were completed at the participants' own pace. The lack of comparison regarding the dosage of education and training provided to the parents prevented the identification of an optimal amount of education and training to achieve the maximum benefit to the children.

The methods of delivery for the parent-mediated interventions were equally as wide-ranging, with Hamad et al. (2010), Heitzman-Powell et al. (2014), and the study by Ingersoll and Berger (2015), Ingersoll et al. (2016), and Pickard et al. (2016), requiring parents to access resources on the Web and progress through the content at their own pace. Heitzman-Powell et al. (2014) coupled the Web-based modules with distant coaching sessions delivered by qualified clinicians. The studies by St. Peter et al. (2014), Wacker et al. (2013), and Vismara et al. (2012) utilized a telehealth delivery model with live, 2-way videoconferencing by qualified clinicians who delivered the intervention in isolation, coupled with a Web-based self-guided website or using teaching materials contained on a DVD. St. Peter et al. (2014) compared the difference between the effectiveness of delivery methods; training provided using video methods versus training provided via a written manual. Finally, the study by Ingersoll and Berger (2015), Ingersoll et al. (2016), and Pickard et al. (2016) compared 2 groups; one receiving access to a Web-based training



program only and the other having access to the same Web-based training program, but with additional weekly therapist-assistance via videoconferencing.

Identifying the superior delivery method of intervention for this population is limited by a lack of between-group comparisons within the included studies. Only the studies by Ingersoll and Berger (2015), Ingersoll et al. (2016) and Pickard et al. (2016), and St. Peter et al. (2014) had comparison groups. Methods with increased user interaction demonstrated some superiority with DVDs having higher adherence to the training program compared with written content. Furthermore, regular therapist-assisted sessions resulted in increased intervention completion, parent appropriateness of intervention, and improvements in parent knowledge and skills.

Overall, these findings suggest that training delivered to parents who live outside of urban areas or with limited access to services can have some effect in improving the social behavior and communication skills in their child with ASD and a large effect on increasing their own knowledge and skills in of ASD interventions. Additionally, no specific content or dosage can be identified as being superior; however, more interactive methods of delivery, such as videos and regular therapist contact for training have been proven to (1) improve adherence, (2) increase completion rates, and (3) improve fidelity in parent-mediated interventions.

#### **2.4.7 Risk of bias in included studies**

The St. Peter et al. (2014), Ingersoll and Berger (2015), Ingersoll et al. (2016), and Pickard et al. (2016) studies assigned participants to different intervention groups. The remaining 5 articles have a high risk of selection bias. In the study by St. Peter et al. (2014), the randomisation process was poorly described with no mention of blinding and allocation procedures by the researchers. The authors reported homogeneity between samples with no

significant differences in socioeconomic status, educational level, or previous experience with the intervention between the experimental and control groups and autism severity scores. Therefore, the risk of bias from confounding variables was reduced due to the homogeneity of the 2 groups. Confounding bias was addressed in the study by Ingersoll and Berger (2015), Ingersoll et al. (2016), and Pickard et al. (2016) by matching participants on their pretreatment expressive language age using a standardized assessment prior to randomisation.

All 7 studies were subject to a high risk of bias due to a lack of blinding. Five of the studies in this review were at a higher risk of confounding bias due to the lack of controls. The small sample sizes of these articles increased the likelihood of type II errors with no article reporting a power calculation relative to the outcome measures.

## **2.5 Discussion**

### **2.5.1 Principal findings**

Findings of this systematic review provide preliminary evidence that parent-mediated intervention training for families living in nonurban areas can assist in improving social behavior and communication skills of children with ASD. Weak study design, lack of standardized outcome measures, lack of measurement outcomes in children with ASD, small participant numbers, high risk of bias, and large variations in interventions limit the generalizability and conclusiveness of the findings to the target population. Despite the limitations, preliminary findings from this review suggest that parent-mediated intervention training delivered remotely could benefit both parents and children with ASD given the barriers they face in accessing traditional services.

The notion that parent-mediated interventions can fully address the gap of limited access to services and be an effective alternative intervention for children with ASD needs further

investigation. A systematic review conducted by McConachie and Diggle (2007) focused on parent-delivered interventions regardless of geographical location or method of delivery for children with ASD. The authors concluded that whereas these types of interventions can improve the social behavior and communication challenges of children with ASD, the lack of studies with robust study design limits the ability to draw further conclusions and highlighted the need for further research. This paper builds on these findings by reviewing current literature on the effectiveness of parent-mediated intervention training delivered remotely to a nonurban population who face a number of barriers accessing traditional services.

In this review, effect sizes were larger for intervention outcomes that targeted parents' knowledge and intervention fidelity skills, compared with intervention outcomes to improve social behavior and communication skills for their children. Only 2 studies included measures of social behavior and communication skills in the children with ASD despite all the interventions providing training for parents to deliver therapy to address these skills. This finding indicates that parents have the potential to improve their knowledge and intervention fidelity skills and be agents in the delivery of therapeutic interventions, thereby improving the social behavior and communication skills of their children with ASD.

The results of this review indicate that the use of telehealth, Web-based modules, and DVDs all seem to have some effect in educating parents about ASD and increasing the fidelity in the delivery of interventions. A lack of standardized measurements and RCTs limited the comparison of interventions within this review. Interventions that were delivered using videos were more effective and accepted by parents than written information. Additionally, weekly contact with a therapist to answer questions and provide

coaching proved to be more effective in the areas of (1) intervention appropriateness, (2) program completion, (3) parent intervention fidelity, (4) parent engagement, and (5) parent's positive perception of their child, when compared to a self-directed program alone. Considering this, the interventions created for families that have limited access to face-to-face therapy could be tailored to meet the needs of the individual parents based on their proximity to services, personal qualities, resources, and preference. Furthermore, interventions clearly benefitted from regular contact with trained professionals throughout the training program.

Defining populations based on their geographical location is challenging due to differing methodologies and definitions adopted by different countries. This disparity in terminology and classification systems makes trying to understand the unique characteristics of families having a child with ASD and living in regional and remote areas difficult due to the wide variability of proximity and access to appropriate services. This is confounded when trying to compare populations from different countries that use vastly different classification systems. The review highlights the importance for researchers to use the relevant geographical classification system in their country to make defining study populations more clearly thereby providing better context for their study.

Finally, evidence is emerging that suggests there is indeed a significant difference in the characteristics and needs of families having a child with ASD residing in urban areas and those residing in rural areas, but further investigation is needed (Hoogsteen & Woodgate, 2013a; Hoogsteen & Woodgate, 2013c; Hutton & Caron, 2005; Murphy & Ruble, 2012). Intuitively this discrepancy between the populations makes sense; however, the poor description of participant characteristics, lack of control groups, and absence of comparisons between these 2 groups prevent conclusive findings.

### **2.5.2 Recommendations for future research**

Further research into the feasibility, efficacy, and appropriateness of the methods of delivery for this unique population will help inform clinical decisions. This systematic review provides preliminary evidence on the effectiveness of remotely delivered parent-mediated intervention training. However, more research is needed to determine the most effective balance between parent-mediated intervention and therapist support via Web-based or distance training to provide the best outcome for a child with ASD, while considering the family's proximity to traditional services. Furthermore, investigation into the effectiveness of the parent-mediated intervention training should not only measure parents' knowledge and skill attainment but also the intervention effectiveness in improving social behavior and communication skills of children with ASD.

Future experimental studies on the effectiveness of parent-mediated interventions, including training programs, should include (1) larger sample sizes, (2) RCTs, (3) improved controls for bias, and (4) use of standardized outcome measures. A lack of comparison groups prevented a meta-analysis in this review. Standardized outcome measures should be employed wherever possible, as these were seldom used in the included studies in this review, with nonvalidated measures often created by the researchers to evaluate the effectiveness of their own intervention. This increased the risk of bias in the studies, thus limiting the impact of the studies' findings. Further research could be focused on comparing different parent training interventions, their components, dosage, and the methods of delivery to determine a superior strategy in increasing parent knowledge and intervention fidelity while improving social behavior and communication skills of their children with ASD.

Despite the studies reporting on the parents' perceived appropriateness and overall

satisfaction of the intervention, there was limited investigation into the influences of parent engagement in the parent-mediated interventions. Further research in relation to the factors surrounding parent engagement in the intervention could help inform clinicians when devising training interventions related to content, parent commitment, and methods of delivery.

There is emerging evidence that interventions delivered remotely can improve the socioemotional and communication skills of children with ASD and may be an alternative to traditional models of therapy (Ramdoss et al., 2011; Ramdoss et al., 2012). The appropriateness and feasibility for parents to utilize other methods to deliver therapy to their children such as direct one-on-one interventions using telehealth technology or the ever-expanding suite of tablet and other ICT-based interventions remains to be comprehensively investigated. Finally, economic modeling comparing the expense of a variety of methods of delivery and interventions could help inform the most cost-effective and feasible delivery method.

The unique context in which families having children with ASD and living in nonurban settings needs to be further researched. Emerging evidence suggests that the nonurban context is different, yet, the unique enablers and barriers in relation to service delivery that these families experience are yet to be fully understood. Furthermore, there is a need for comparison studies between urban and nonurban populations to better develop effective, appropriate, and feasible interventions to improve the social behavior and communication skills in children with ASD; thus allowing the development of tailor-made interventions for each population.

### **2.5.3 Limitations**

A rigorous process involving (1) the searching of 5 databases, (2) establishing interrater

reliability between 2 independent researchers for inclusion or exclusion agreements, (3) standardized data extraction forms, and (4) methodological assessment using the Kmet appraisal checklist was conducted in this study. Despite this, there are some limitations in the review that should be noted. Defining the population was challenging given the poor use of standardized geographical classification systems by authors. Inclusion was based on author report of the participants living in areas described as nonurban, rural, or remote, and as having limited access to services. This could have led to some studies being excluded if this description was not provided by the authors. Additionally, the small number of articles included limits the generalizability of findings to the target population.

## **2.6 Conclusion**

Overall, there is preliminary evidence that parent-mediated intervention training delivered remotely can improve parents' knowledge in ASD, parent intervention fidelity, and subsequently improve the social behavior and communication skills of their children with ASD. The studies included in this review had an unclear or high risk of bias due to a lack of control groups and paucity of using standardized outcome measures. Additionally, difficulties in defining the participant characteristics limited the translatability to the target population. Few studies reported on the feasibility and appropriateness of the interventions and the factors of parent engagement in the interventions were evident in most studies. Future research should aim to use RCT designs, incorporate standardized outcome measures, and describe participant characteristics in greater detail. Furthermore, the review highlighted the need to investigate the feasibility and appropriateness of the interventions in addition to the factors influencing parent engagement in the interventions.

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## Chapter 3      Stress, Coping and Quality of Life in parents of children with ASD living regionally

Chapter 3 describes findings from a cross-sectional survey undertaken to explore whether regionality is associated with differing stress levels, coping, quality of life and daily routines for parents and families of a child with autism spectrum disorder (aged 2 - 18 years) in Western Australia. The survey used validated outcome measures and compared the stress levels and QoL of this group to population norms. This chapter has been accepted and is currently in press in the Journal of Child and Family Studies.

The spelling and wording contained within this chapter are that of the submitted manuscript.

Note: The terms low densely populated (LDP) and densely populated (DP) areas are used in this manuscript. Both LDP and DP areas comprise of remoteness areas from the Australian Standard Geographical Classification (ASGC). LDP areas comprise the remoteness areas *very remote*, *remote* and *outer regional*, with *inner regional* and *major cities* being group together as DP areas.





# Journal Manuscript 2

## **Title**

Stress, coping, and quality of life in families with a child with ASD living regionally

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### 3.1 Abstract

**Objectives:** The present study aimed to explore whether regionality is associated with differing stress levels, coping, QoL and daily routines for parents and families of a child with ASD (aged 2 - 18 years) in Western Australia using validated outcome measures and compare the stress levels and QoL of this group to population norms.

**Methods:** A sample of 278 families living in Western Australia who have a child or adolescent (2-18 years old) with a clinical diagnosis of ASD participated in a cross-sectional survey. Multivariate logistic regression modelling was conducted to determine key factors associated between regionality and demographic variables, quality of life, coping styles, time use, and stress levels.

**Results:** Parents living in low densely populated areas were more likely to adopt avoidant coping mechanisms, compared to those living in densely populated areas. Fathers with children on the autism spectrum were less likely to be educated above diploma level in regional and remote areas. Stress, QoL or daily routines did not differ by regionality; however, the total sample (i.e., parents from both LDP and DP areas) experienced significantly higher levels of stress and lower QoL when compared to the general population.

**Conclusion:** The findings suggest that despite having higher levels of stress and lower QoL compared to the general population, residing in a geographically LDP area in Western Australia has a small association on preferred coping style preference and has no association on stress levels, QoL or daily routines for parents who have a child with ASD.

**Keywords:** Autism Spectrum Disorder, Quality of Life, Parents, Psychological Adaptation



### **3.2 Introduction**

Autism Spectrum Disorders (ASD) include a life-long spectrum of conditions characterised by deficits in social communication and behaviour, including problems interpreting non-verbal gestures, difficulty developing age-appropriate friendships, rigid adherence to routines, and adapting to environmental change (American Psychiatric Association, 2013; Happé & Ronald, 2008). In the absence of pharmaceutical intervention, intensive early intervention delivered by trained clinicians is recommended to ameliorate core symptomology and thereby improve functional outcomes (Dawson et al., 2010; Whalen, Liden, Ingersoll, Dallaire, & Liden, 2006). The symptoms and concomitant challenging behaviours associated with ASD present parents and children with many adverse outcomes, including but not limited to, higher parental stress, poor sibling adjustment, family functioning, disruptive behaviour, and social isolation (Gray, 1994; Rao & Beidel, 2009).

Parents of a child with ASD experience higher levels of stress compared to parents of typically developing children, as well as parents of children with other disabilities (Dabrowska & Pisula, 2010; Duarte, Bordin, Yazigi, & Mooney, 2005; Hayes & Watson, 2013; Higgins, Bailey, & Pearce, 2005; Schieve, Blumberg, Rice, Visser, & Boyle, 2007). Frequent contributors to parental stress in ASD include, social behaviour challenges of children with ASD, parents' reduced ability to socialise, reduced access to individual therapy, negative co-parent relationships and high out of pocket costs (Horlin, Falkmer, Parsons, Albrecht, & Falkmer, 2014; Johnson, Frenn, Feetham, & Simpson, 2011; Lecavalier, Leone, & Wiltz, 2006; Sim et al., 2018). Higher levels of stress have also been found to be a predictor of lower quality of life (QoL) (Khanna et al., 2011; Lee et al., 2009). A recent systematic review by Vasilopoulou and Nisbet (2016) concluded that parents of children with ASD are more likely to experience poorer QoL when compared with parents of typically developing children or to population norms. The authors

concluded that variables associated with lower parental QoL were child behavioural difficulties, unemployment, being a mother and a lack of social support. Furthermore, the QoL of parents with a child with ASD has been shown to directly impact the QoL of their children, with lower QoL experienced by parents associated with lower QoL in their children (Burgess & Gutstein, 2007). Given the bi-directional nature of the parent-child relationship, consideration of the QoL of parents is an essential factor when working with this population.

The transactional model of stress and coping proposed by Lazarus and Folkman (1984) viewed stress as the outcome of the interaction between the environment and the individual. When environmental stressors/demands exceed individual resources, coping mechanisms are recruited to restore function. According to Lazarus and Folkman (1984), if the coping mechanisms cannot meet the demands or are maladaptive, the outcome is stress. Subsequently, if stress is the outcome of inadequate coping mechanisms, it would be remiss not to investigate individual preferences for coping in parents of children with ASD.

Parents of children with ASD utilise a range of coping strategies when stressed and the use of these coping strategies are variable compared to parents of typically developing children or children with other disabilities (Hastings et al., 2005; Lai, Goh, Oei, & Sung, 2015).

Hastings et al. (2005) explored the structure of coping strategies used by parents of children with ASD living in the United Kingdom. The authors reported four distinct coping dimensions: 1) active avoidance coping, such as self-blame; 2) problem-focused coping, such as planning and taking action to address the problem; 3) positive coping, such as humour or positive reframing; and 4) religious/denial coping, such as prayer. A review paper by Lai and Oei (2014) reported that parents of children with ASD recruit both adaptive (seeking social support and problem-focused) and maladaptive (active-avoidance)

coping strategies to manage stress related to caregiving. Furthermore, Lai and Oei (2014) concluded that maladaptive coping strategies contributed to higher levels of stress and mental health problems, while adaptive coping strategies are associated with lower levels of stress and fewer mental health problems - such as depression. A gap remains in the research regarding the nature of parental coping in ASD across different individuals and situations. Specifically, broad coping frameworks for parents with ASD may not be generalisable to all parents and caregivers of children with ASD from different cultural, demographic and environmental contexts.

Carver, Scheier, and Weintraub (1989) suggested that coping is context-dependent and primarily influenced by the constraints of the situation. The unique context of living in regional and remote Australia and the comparison between parents living in these areas and those living in more densely populated areas in relation to their stress levels is poorly understood and further investigation is warranted. In this paper, regionality is defined in terms of the difference in population density. Terms of densely populated (DP) and low densely populated (LDP) are used to differentiate between highly populated areas and regional and remote areas, respectively (McAuliffe, Vaz, Falkmer, & Cordier, 2016).

The challenges of raising a child with ASD can be magnified for families living in regional and remote areas in Australia, where access to timely and appropriate health and therapeutic services can potentially create unique barriers (Antezana, Scarpa, Valdespino, Albright, & Richey, 2017; Chen, Liu, Su, Huang, & Lin, 2008; Hutton & Caron, 2005; Murphy & Ruble, 2012), which, in turn, may impact on the type of coping strategies used by parents. Specifically, families of children with ASD living in regional and remote areas in Australia attempting to access appropriate health services can encounter: 1) widely dispersed services requiring parents to travel vast distances to access services; 2) lower

levels of screening and delayed diagnosis; and 3) difficulties in retaining skilled health professionals in these settings (Iacono, Humphreys, Davis, & Chandler, 2004; Smith, Humphreys, & Wilson, 2008; Struber, 2004). These are unique stressors to families living in LDP areas, which could negatively influence stress levels and coping strategies adopted by parents living in these areas (Elgar, Arlett, & Groves, 2003). Moreover, there is a scarcity of literature investigating the difference in stress levels and coping strategies between families living in DP areas compared to those living in LDP, even in families who do not have a child with ASD. Given the importance of early intervention in reducing the child's ASD related behaviours; limited access to adequate health services and a shortage of adequately trained early intervention health and education professionals are of particular concern for this population, serving to increase parental stress (Hayes & Watson, 2013; Hutton & Caron, 2005; Iacono et al., 2004; Smith et al., 2008; Struber, 2004).

The unique nature of stress and the interaction between stressors and the environment of parents of children with ASD living in regional and remote areas in Australia could affect the coping strategies recruited by this population (Hastings et al., 2005; Hoogsteen & Woodgate, 2013b). Furthermore, there is evidence to suggest that coping strategies used by parents of children with ASD could be different when compared to parents of typically developing children or children with other disabilities, due to the differences in social environments (e.g., poorer social supports and limited access to service) often experienced by this group (Hastings et al., 2005; Lai et al., 2015). Few studies have defined and categorised different types of coping strategies used by parents who have a child with ASD (Benson, 2010; Hastings et al., 2005; McAuliffe, Cordier, Vaz, Thomas, & Falkmer, 2017) and none have done so with families living in regional or remote areas in the Australian context. It is plausible that people living in regional areas in Australia may exhibit different coping mechanisms due to the unique context in which they live, such as limited social



support and poorer access to services. However, an extensive literature search indicates a scarcity of research having been undertaken to investigate this.

Synthesising the findings from studies that investigated the impact of regionality on study outcomes is, however, a challenge, due to heterogeneity in classification systems, geographical topographies, poor sampling methods within studies and differences in local government policy and resourcing (Hoogsteen & Woodgate, 2013b; Murphy & Ruble, 2012; Parsons, Cordier, Vaz, & Lee, 2017). To date, only one empirical study has explicitly investigated the role of regionality in Australia in the lives of parents of children with ASD. McAuliffe et al. (2016) compared family daily routines, service usage and stress levels of parents of children with ASD living across Western Australia by regionality and concluded that families living in low densely populated areas experience reduced employment hours, travel greater distances to access medical facilities and report less severe stress levels when compared to families living in urban areas. However, McAuliffe et al. (2016) did not use validated assessments to measure the study outcomes and did not investigate the phenomena of coping or QoL. The current study extends the knowledge base by: 1) Exploring whether regionality is associated with differing stress levels, coping, QoL and daily routines for parents and families of a child with ASD (aged 2 - 18 years) in Western Australia using validated outcome measures; and 2) Comparing stress levels and QoL of parents and families of a child with ASD (aged 2 - 18 years) in Western Australia with population norms.

### **3.3 Methods**

#### **3.3.1 Participants**

Families living in WA who had a child or adolescent (2 - 18 years old) with a clinical diagnosis of ASD determined by a team of qualified health professionals using the DSM-5

or DSM-IV criteria (if diagnosed prior to the DSM-5) were recruited (American Psychiatric Association, 2013). Families were recruited through the Disability Services Commission (DSC) of WA by contacting every parent of children with ASD on their register; by the research team contacting service providers, such as general practitioners, paediatricians, speech pathologist and occupational therapists working with families of children with ASD; related events run through Curtin University, community organisations, such as the Southwest Autism Group (SWAN); and ASD service providers in WA. A list of families who have agreed to be contacted for research studies, housed at Curtin University and the Telethon Kids Institute, were also invited to participate.

Survey responses from 278 families of children with ASD were received, with 91% of respondents ( $n = 255$ ) female and 9% ( $n = 24$ ) male. The characteristics of the families who responded to the survey are summarised in Table 3.1. There were no significant differences between families living in DP areas to those living in LDP areas except for the father's education in the family, with fathers in LDP having lower education levels compared to fathers living in DP areas. The characteristics of the children with ASD collected in the survey are displayed in Table 3.2. The age when the children first accessed therapeutic services was the only significant difference between those children living in DP areas and those living in LDP areas.

**Table 3.1 Characteristics of families of children with ASD living in Densely Populated (DP) areas vs Low Densely Populated (LDP) areas**

Characteristics	Total sample		Living in DP areas		Living in LDP areas	
	N	%	N	%	N	%
<b>Total</b>	278	100	230	82.1	48	17.1
<b>Number of children with ASD</b>						
1	238	85.9	200	87.3	38	79.2
More than 1	39	14.1	29	12.7	10	20.8
$\chi^2 = 2.20, df = 1, p = .139$						
<b>Household comp</b>						
Two parent	195	74.4	159	73.6	36	78.3
Single	48	18.3	40	18.5	8	17.4
Other	19	7.3	17	7.9	2	4.3
$\chi^2 = .785, df = 2, p = .675$						
<b>Total number of children</b>						
1	39	14.9	36	16.7	3	6.7
2 or more children	222	85.1	180	83.3	42	93.3
$\chi^2 = 2.93; df = 1, p = .087$						
<b>Mother's education</b>						
Up to diploma	159	60.9	131	60.6	28	62.2
Undergraduate Degree or higher	102	39.1	85	39.4	17	37.8
$\chi^2 = .039; df = 1, p = .844$						
<b>Father's education</b>						
Up to diploma	170	69.1	132	65.3	38	86.4
Undergraduate Degree or higher	76	30.9	70	34.7	6	13.6
$\chi^2 = 7.475; df = 1, p = .006^{**}$						
<b>Employment status of household</b>						
Employed	211	89	171	89.1	40	88.9
Unemployed	26	11	21	10.9	5	11.1
$\chi^2 = .001, df = 1, p = .973$						
<b>Household Income<sup>1</sup></b>						
Up to \$51,999	31	13.9	26	14.4	5	11.9
\$52,000 and over	192	86.1	155	85.6	37	88.1
$\chi^2 = 1.72, df = 1, p = .678$						

<sup>1</sup>The cut-off point was set based on the median household income in Western Australia at the time of the study

\*Significant result  $p < .05$

\*\*Significant results  $p < .01$

**Table 3.2 Characteristics of children with ASD living in Densely Populated (DP) areas vs. Low Densely Populated (LDP) areas**

Characteristics	Total sample		Living in DP areas		Living in LDP areas	
	N=	%	N=	%	N=	%
<b>Children’s Age (month):</b>						
Mean (standard deviation)	116.5 (54.9)		116.4 (54.0)		115.2 (58.9)	
	<i>t</i> = .184, <i>p</i> = .85					
<b>Gender</b>						
Boy	230	82.7	192	83.5	38	79.2
Girl	48	17.3	38	16.5	10	20.8
	<i>X</i> <sup>2</sup> = .52, df = 1, <i>p</i> = .47					
<b>Age when first sign of ‘something not right’ noticed</b>						
Less than 3 years old	225	80.9	187	81.3	38	79.2
3 years old and older	53	19.1	43	18.7	10	20.8
	<i>X</i> <sup>2</sup> = .52, df = 1, <i>p</i> = .47					
<b>Age when formally diagnosed</b>						
Less than 4 years old	119	62.2	99	43.2	20	42.6
Between 4 and 6 years old	70	19.4	58	25.3	12	25.5
Older than 6 years old	87	18.3	72	31.4	15	31.9
	<i>X</i> <sup>2</sup> = .01 <i>df</i> = 2, <i>p</i> = 1.00					
<b>Age when therapy first accessed</b>						
Less than 3 years old	82	29.6	71	30.9	11	23.4
Between 3-4 years old	72	26.0	52	22.6	20	42.6
Between 4-6 years old	71	25.6	63	27.4	8	17.0
Older than 6 years old	52	18.8	44	19.1	8	17.0
	<i>X</i> <sup>2</sup> = 8.44, <i>df</i> = 3, <i>p</i> = .038*					

\*Significant result  $p < .05$

\*\*Significant result  $p < .01$

### **3.3.2 Procedure**

A cross-sectional survey design was used. Ethics approval was received from the Curtin University Human Ethics Committee (HR123/2014) for this study. Potential participants had one of three possible options to participate in the survey: 1) online, 2) via telephone, or 3) using pencil and paper. The online survey was available from January 2015 to December 2016. Consent for the online version was provided by ticking a box at the start of the survey. If participants elected to complete the survey over the telephone, a member of the research team would call at a nominated time convenient for the participant. Pre-determined inclusion criteria for all methods of completing the survey were established prior to commencing the survey. The inclusion criteria were having a child or adolescent (2 - 18 years old) with ASD; the child having no other major medical or psychiatric conditions (e.g., Fragile X syndrome); and the family resided in Western Australia (WA). Once consent to participate was obtained and the participant deemed eligible, the researcher administered the survey. Finally, if the participants elected to complete the survey using the paper and pencil method, a pack was sent in the post; including a consent form to be returned via an enclosed envelope separate to the survey. All data whether entered directly by participants online or by researchers from the paper copies were housed on password protected Curtin University's Qualtrics Web Server.

### **3.3.3 Measures**

To create the survey, the authors undertook a literature search of studies investigating the lived experience of living regionality with ASD, researched measures with robust psychometrics for this population, and incorporated feedback from representatives of local government and support groups. Survey data were collected on: 1) ASD identification and diagnostic procedures; 2) availability and accessibility of services and support; 3) parent satisfaction with services and supports; 4) direct financial costs of parenting a child with

ASD; and 5) emotional stress and physical barriers to receiving professional input. The following validated instruments were incorporated: Autism Parenting Stress Index (APSI) (Silva & Schalock, 2012a, 2012b); Brief COPE (Carver, 1997); and World Health Organisation Quality Of Life - BREF (Skevington, Lotfy, & O'Connell, 2004).

#### **3.3.3.1 Autism Parenting Stress Index**

The Autism Parenting Stress Index (APSI) is a valid and reliable instrument for measuring parenting stress in relation to the core and co-morbid symptoms of ASD (Silva et al., 2015). The measure is intended for use by clinicians to identify areas where parents need support with parenting skills and to assess the effect of the intervention on parenting stress (Silva & Schalock, 2012b). A validation study of the measure reported internal consistency for parents of children with ASD was acceptable with Cronbach's  $\alpha$  scores .76, .76 and .67 on the factors of core autism behaviours, co-morbid behaviours and co-morbid physical issues. Test-retest reliability coefficient was .88 at a 4-month interval (Silva & Schalock, 2012b; Silva, Schalock, & Gabrielsen, 2011). The APSI uses a five-point Likert scale and respondents are asked to rate aspects of their child's health according to how much stress it causes them or their family. Some example items include "Your child's ability to communicate" and "Difficulty making transitions from one activity to another" (Silva & Schalock, 2012b)

#### **3.3.3.2 Brief COPE**

The Brief COPE is an abbreviated version of the COPE inventory. The Brief COPE uses a 4-point Likert scale to determine how frequently they employ 28 different behaviours and cognitions across 14 scales when coping with stressful situations (Benson, 2010; Carver, 1997). Although not specific to measuring stress in the context of parenting children with ASD, the Brief COPE is a useful tool in identifying poor coping outcomes and has been

used extensively as an outcome measure with this population (Benson, 2010; Hastings et al., 2005; Lai & Oei, 2014). Except for three scales, Venting (.50), Denial (.54) and Acceptance (.57), Cronbach's  $\alpha$  scores are an average of .72 (range .52 - .93) across the 14 scales (Benson, 2010). Some example items include "I've been taking action to try to make the situation better" and "I've been getting emotional support from others" (Carver, 1997).

### **3.3.3.3 World Health Organisation's Quality of Life- BREF (WHOQOL-BREF)**

The WHOQOL-BREF is an abbreviated 26-item version of the WHOQOL-100 assessment clustered into four domains: 1) physical health; 2) psychological; 3) social relationships; and 4) environment (Skevington et al., 2004). The measure consists of quality of life items that are concerned with the meaning of different aspects of life to the respondents, and how satisfactory or problematic their experience is of them. The assessment covers a broad range of facets and has been cross-culturally validated for several languages (Skevington et al., 2004). Internal consistency for the total sample is acceptable (Cronbach's  $\alpha > .70$ ) for three out of the four domains for physical health (.82), psychological (.81), and environment (.80), but marginal for the social relationships domain (.68). Test-retest reliability is generally high ranging from .56 to .84 for individual items over an interval from 2-8 weeks. Domain test-retest are .66 for physical health, .72 for psychological, .76 for social relationship and .87 for the environment domains (Skevington et al., 2004; WHOQOL Group, 1998). The measure's domain scores have been shown to correlate at approximately .90 with the WHOQOL-100 domain scores, which has itself demonstrated sound criterion validity. The domain scores have also been shown to demonstrate content and discriminate validity (WHOQOL Group, 1998). Some example items include "To what extent do you feel that physical pain prevents you from doing what you need to do?" and "How would you rate your quality of life?". (WHOQOL Group, 1998).

#### **3.3.3.4 Daily routines**

Participants were asked to complete a 24-hour format table to describe their average weekday and weekend day by providing estimated hours on each of the 14 activities presented. These 14 activities were chosen based on published literature investigating daily routines of parents of children with a disability (Gevir, Goldstand, Weintraub, & Parush, 2006; McCann, Bull, & Winzenberg, 2012). The 14 activities listed in this study were: 1) sleep; 2) grooming and personal hygiene; 3) meal preparations and clean up; 4) having a meal; 5) taking care of child; 6) travel time to and from school or work; 7) time spent at work; 8) personal leisure time; 9) house duties; 10) shopping; 11) having quiet time or down time; 12) visiting family/ friends; 13) studying; and 14) voluntary work.

#### **3.3.4 Data analyses**

Data were managed and analysed using IBM SPSS<sup>®</sup> Statistics version 24 (IBM Corp., 2016). Descriptive statistics were used to describe the demographic characteristics of families living in low densely populated (LDP) and families living in densely populated (DP) areas. Independent *t*-tests for continuous variables and Pearson's  $\chi^2$  for categorical variables were performed to determine between-group differences using the dependent variable of regionality with the independent variables of key child factors (e.g., age, diagnosis, comorbidity), and parent factors (e.g., demographic factors, stress, coping, quality of life, impact on routine). Binary logistic regression analysis was conducted on significant variables using the backward elimination method to develop the final model to determine key factors associated with demographic variables, quality of life, coping styles, time use, and stress levels, using the dependent variable of regionality. Finally, unequal variance *t*-tests were performed to compare the total sample (DP and LDP combined) with published general population normative data for the WHOQOL-BREF and APSI outcome measures (Hawthorne, Herrman, & Murphy, 2006; Silva & Schalock, 2012b).



For this study, regionality was defined using the Australian Bureau of Statistics (ABS) five-category classification based on the Australian Standard Geographical Remoteness Classification System (ASGC) (Australian Bureau of Statistics, 2011a). The aim of ASGC remoteness structure is to divide Australia into broad regions for comparative statistical purposes. There is no widely accepted standard to determine when city becomes country; the ASGC remoteness classification system simply groups areas where all members of that remoteness area (RAs) have similar, but not identical, characteristics of remoteness (Australian Bureau of Statistics, 2003). The ASGC remoteness structure is classified into five RAs (Australian Bureau of Statistics, 2011a). The categories include: 1) major cities; 2) inner regional; 3) outer regional; 4) remote; and 5) very remote. RAs are based on road distances to the nearest service centres, and average scores are calculated using the Accessibility/Remoteness Index of Australia (ARIA +) grid, which is a one square kilometre grid covering all of Australia (Australian Bureau of Statistics, 2011b; McAuliffe et al., 2016). Participants' postcodes in the current study were recoded based on this classification.

The RAs were further collapsed into a dichotomous variable due to the small number of respondents from remote and very remote areas. In doing so, the DP category ( $n = 230$ , 82.7%) was formed by collapsing the major city ( $n = 136$ , 48.9%) and inner regional city ( $n = 94$ , 33.8%) postcodes and LDP category ( $n = 48$ , 17.3%) was formed by collapsing the outer regional ( $n = 29$ , 10.4%), remote ( $n = 15$ , 5.4%) and very remote postcodes ( $n = 4$ , 1.4%). The rationale for this dichotomisation was based on people living in outer regional, remote and very remote areas having to travel significantly greater distances to access services compared to those living in major cities and inner regional areas (Australian Bureau of Statistics, 2011b). Analyses were also conducted with the DP category ( $n = 136$ , 48.9%) comprising only of participants from major cities and the LDP category ( $n = 142$ ,

51.1%) consisting of participants from inner regional, outer regional, remote and very remote regions. No difference in findings was observed, regardless if inner regional data were dichotomised with DP or LDP categories.

An exploratory factor analysis was conducted to best fit the current study's participants (see Table 3.3) as the studies by Benson (2010) ( $n = 113$ ) and Hastings et al. (2005) had smaller sample sizes ( $n = 113$ ;  $n = 135$  respectively) than the current study ( $n = 278$ ) and did not contain such a large proportion of participants from regional and remote areas. A principle component analysis using an oblimin rotation was completed for the 28 items used in the measure. The scree-test was used to determine the number of factors with the eigenvalue greater than 1.0 (Field, 2013; Osborne & Costello, 2009) and factor loading greater than .40. Following the initial examination of the correlation matrix, the two items, which form a subscale of substance use and another two items, which form a subscale of use of religion, were highly correlated ( $r > .8$ ) and therefore excluded. Eigenvalues greater than two indicated that the first three factors explained 20%, 15%, and 8%, of the variance, respectively (Table 3.3). The fourth, fifth and sixth factors had eigenvalues under 2, explaining 8%, 5% and 5% of the variance, respectively. The scree plot was ambiguous showing inflections on factors three and five, justifying retaining both of these factors; however, the components did not fit the theoretical framework validated by Hastings et al. (2005) or Benson (2010), hence were excluded. The Kaiser-Meyer-Olkein measure indicated good sampling adequacy ( $KMO = .768$ ) and Bartlett's test of sphericity ( $\chi^2 = 2213.23$ ,  $df = 276$ ,  $p < .001$ ) indicated sufficient correlation between each item (Field, 2013). As a result, the three-factor solution explaining 43% of the total variance was used in the current study. Cronbach's  $\alpha$ -values are reported for all three factors in Table 3.3. In contrast to the studies by Benson (2010) and Hastings et al. (2005) – each had four factors – our analysis yielded three factors with one item dropped: 1) problem-focused; 2) active

avoidance; and 3) positive coping. The item dropped was an item for the self-distraction subscale, *'I have been turning to work or other activities to take mind off things'*.

Due to the similarities between our extracted factors and those of Hastings et al. (2005) with the exception of religious/denial coping, we elected to use their factor labels in this study. The problem-focused factor refers to the proactive coping styles to deal with stressors, including items of the planning, use of instrumental support, active coping, and acceptance subscales. Active avoidance described coping styles that avoided physical and emotional stressors, including items of self-blame, behavioural disengagement, denial and venting in addition to one item from the self-distraction subscale. Finally, positive coping described coping by perceiving stressors through a constructive lens. This included items of the humour, positive reframing and use of emotional support subscales.

**Table 3.3 Factor loadings for analysis of Brief COPE items**

Factor	Problem focused	Active avoidance	Positive coping	Limited use of relationship support	Acceptance
% variance	18.44	15.34	9.02	7.74	5.74
Cronbach's alpha	.77	.78	.74	.75	.70
Coping taking action to try to make the situation better	<b>.722</b>	-.052	.028	-.001	.172
Coping concentrating my efforts on doing something about the situation I am in	<b>.700</b>	.081	.033	-.068	-.087
Coping trying to come up with a strategy about what to do	<b>.652</b>	.208	.041	-.046	.244
Coping thinking hard about what steps to take	<b>.578</b>	.117	-.036	-.028	<b>.450</b>
Coping blaming myself for things that happened	.147	<b>.738</b>	-.009	.028	-.035
Coping giving up the attempt to cope	-.205	<b>.735</b>	-.184	.048	.186
Coping criticizing myself	.151	<b>.721</b>	-.002	.119	-.102
Coping saying to myself this isn't real	.096	<b>.646</b>	.131	.120	-.340
Coping refusing to believe that it has happened	.231	<b>.615</b>	-.011	.040	<b>-.414</b>
Coping giving up trying to deal with it	<b>-.470</b>	<b>.543</b>	-.017	.109	.335
Coping saying things to let my unpleasant feelings escape	-.076	<b>.523</b>	-.122	-.370	-.123
Coping doing something to think about it less, such as going to movies, watching TV, reading, daydreaming, sleeping, or shopping	-.260	<b>.409</b>	.233	-.245	.188
Coping turning to work or other activities to take my mind off things	.010	.349	.090	-.023	.172
Coping making jokes about it	-.080	.009	<b>.894</b>	.042	-.039
Coping making fun of the situation	-.161	.009	<b>.861</b>	.001	-.134
Coping trying to see it in a different light, to make it seem more positive	.325	.069	<b>.594</b>	-.050	.127
Coping looking for something good in what is happening	.211	-.150	<b>.536</b>	-.083	.195
Coping getting emotional support from others	-.075	-.058	-.047	<b>-.836</b>	.015
Coping getting comfort and understanding from someone	-.043	-.231	.139	<b>-.726</b>	-.029
Coping getting help and advice from other people	.366	-.199	.100	<b>-.611</b>	-.064

Factor	Problem focused	Active avoidance	Positive coping	Limited use of relationship support	Acceptance
Coping expressing my negative feelings	-.055	<b>.409</b>	.144	<b>-.570</b>	.002
Coping trying to get advice or help from other people about what to do	.359	.084	-.081	<b>-.566</b>	.040
Coping learning to live with it	.137	-.062	.097	.104	<b>.715</b>
Coping accepting the reality of the fact that it has happened	.234	-.099	.007	-.081	<b>.655</b>

*Note.* Significant loadings are highlighted in bold.

### 3.4 Results

#### 3.4.1 Factors predictive of regionality

Chi-square tests for categorical outcomes and *t*-tests for continuous outcomes were used to determine univariate differences as a function of regionality. A multivariate logistic regression model of factors that significantly predicted living in low-density areas (LDP) was developed by including factors that approached significance at univariate level ( $p < .10$ ). The following factors were included in the multivariate model: 1) number of children with ASD in the family; 2) age the child first accessed therapy; 3) level of fathers education; 4) avoidance coping factor cluster in the Brief COPE; 5) Social domain in the WHOQOL-BREF; 6) Environmental domain in the WHOQOL-BREF; 7) Co-morbid behaviour domain in the Autism Parenting Stress Index; 8) hours sleeping on the weekend; and 9) hours spent studying on the weekend.

The final multivariate model is presented in Table 3.4. Backward elimination was used to build the model as there was no a priori rationale to enter variables into the model (Field, 2013). Goodness of fit of the model was tested against a constant only model and found to be statistically significant ( $\chi^2(5) = 28.58, p < .001$ ). The final model explained 20% of the variability in regionality (Nagelkerke's  $R^2 = .20$ ). The overall accuracy of the model was

83.8%. The Wald criterion and  $Exp(\beta)$  were used to predict the strength of individual factors. Parents who lived in LDP areas were 1.1 times more likely to adopt avoidance coping strategies ( $p = .004$ ,  $Exp(\beta) = 1.14$ ) when compared to parents living in DP areas. Fathers who lived in LDP were 3.4 times more likely to have achieved a qualification up to a diploma or lower ( $p = .017$ ,  $Exp(\beta) = 3.44$ ).

The multivariate logistic regression analysis detected no significant difference for associations between the dependent (regionality) and the following independent variables: 1) number of children with ASD in the family; 2) age the child first accessed therapy; 3) Social domain in the WHOQOL-BREF; 4) Environmental domain in the WHOQOL-BREF; 5) co-morbid behaviour domain in the Autism Parenting Stress Index; 6) hours sleeping on the weekend; and 7) hours spent studying on the weekend. Lastly, sensitivity analyses were conducted by including parent gender, parent's age, child gender, and time since the diagnosis was made as covariates in the model; however, no significant differences in these independent variables were detected and they did not change the final model.

**Table 3.4 Variables associated with living in Low Densely Populated areas.**

Variables	<i>B</i>	SE	Wald	<i>p</i>	<i>Exp</i> ( $\beta$ )	95 % CI for <i>Exp</i> ( $\beta$ )	
						Upper	Lower
<b>Constant</b>	-5.07	1.45	12.20	< .001	< .01		
<b>Number of Children with ASD</b>	-1.02	.59	3.03	.082	.36	.12	1.14
<b>Father's Highest Education Level</b>	1.24	.51	5.69	.017*	3.44	1.24	9.48
<b>Avoidance Coping</b>	.13	.04	8.33	.004**	1.14	1.04	1.24
<b>Acceptance Coping</b>	.17	.01	3.18	.075	1.19	.98	1.43
<b>Hours study weekend</b>	-.68	.40	2.97	.085	.50	.23	1.10

\*Significant result  $p < .05$

\*\*Significant result  $p < .01$

### **3.4.2 Comparison to general population**

Respondent scores from the overall sample (DP and LDP combined) on the WHOQOL-BREF and APSI outcome measures measuring quality of life and stress levels respectively, were compared with published normative data based on the general population (see Table 3.5). Normative data from the WHOQOL-BREF was obtained from a random sample of residents with respondents being 54% female and 46% male, with an average age of 48.2 years (SD = 17.3) living in Victoria, Australia (Hawthorne et al., 2006). Participants were recruited through random telephone selection and stratified including a broad range of health conditions from full health to terminal illness in addition to socioeconomic status (Hawthorne et al., 2006). Normative data for the APSI was obtained from 139 parents of typically developing children completing a survey in Oregon, USA (Silva & Schalock, 2012b). Inclusion criteria for the typically developing children were: 1) between the age of three to six; 2) no diagnosis of ASD; 3) no developmental delay; 4) no chronic illnesses or medical conditions (Silva & Schalock, 2012b). Results demonstrated highly significant differences ( $p < .001$ ) in all domains for both measures. Parents of children with ASD experience four times more stress overall when compared to parents of typically developing children. Furthermore, parents of children experience 14%, 25%, 37% and 18% poorer quality of life compared to parents of typically developing children across the physical, psychological, social and environmental domains of their lives, respectively.

**Table 3.5 Comparison of total sample with general population**

	<b>Total Sample (<i>n</i> = 278)</b>	<b>General Population<sup>1</sup></b>	<b><i>t</i>-score</b>	<b><i>p</i>-value</b>	<b>Cohen's <i>d</i></b>
<b>WHOQOL-BREF Domains<sup>2</sup></b>					
<b>Physical</b>					
Mean	63.13	73.50	-8.61	< .001**	.59
SD	17.26	18.10			
<b>Psychological</b>					
Mean	52.62	70.60	-18.94	< .001**	1.30
SD	13.70	14.00			
<b>Social</b>					
Mean	51.10	81.50	-19.82	< .001**	1.46
SD	23.41	18.20			
<b>Environmental</b>					
Mean	61.56	75.10	-12.86	< .001**	.94
SD	15.93	13.00			
<b>APSI Factors<sup>3</sup></b>					
<b>Stress overall</b>					
Mean	20.84	5.41	20.81	< .001**	-2.04
SD	9.96	5.18			
<b>Core ASD behaviours</b>					
Mean	10.06	1.32	26.58	< .001**	-2.62
SD	4.81	1.86			
<b>Co-morbid behaviours</b>					
Mean	6.16	2.42	12.59	< .001**	-1.24
SD	3.83	2.22			
<b>Co-morbid physical issues</b>					
Mean	5.22	1.67	12.70	< .001**	-1.25
SD	3.54	2.15			

<sup>1</sup>General population sample sizes: WHOQOL–BREF (*n* = 866), APSI (*n* = 139)

<sup>2</sup> Higher scores indicate higher QoL



<sup>3</sup> Lower scores indicate lower levels of stress

\*Significant result  $p < .05$

\*\*Significant results  $p < .01$

### **3.5 Discussion**

The aim of this study was to: 1) explore whether regionality is associated with differing stress levels, coping, QoL and daily routines for parents and families of a child with ASD (aged 2 - 18 years) in Western Australia using validated outcome measures; and 2) compare the stress levels and QoL of this group to population norms. The higher use of avoidant coping strategies and fathers being more likely to have lower education levels were the only significant differences between parents living in LDP areas compared to DP areas. Stress, QoL or daily routines did not differ by regionality; however, the total sample (i.e., parents from both LDP and DP areas) experienced significantly higher levels of stress and lower QoL when compared to the general population. The findings suggest that despite having higher levels of stress and lower QoL compared to the general population, residing in a geographically LDP area in Western Australia has a small association on preferred coping style preference and has no association on stress levels, QoL or daily routines for parents who have a child with ASD.

#### **3.5.1 Coping**

Despite experiencing similar levels of stress and QoL, parents who live in LDP areas were significantly more likely to use avoidant coping strategies compared to those living in DP areas, however, the strength of the association at the group level was small ( $Exp(\beta) = 1.14$ ). Given the lack of research into parents' experiences of having a child with ASD while living in regional Australia, interpretation of this finding presents some challenges. On the one hand, parents from LDP areas may be more likely to adopt avoidant coping

strategies due to poor access to formal support options. On the other hand, the magnitude of the difference between parents living in DP and LDP areas was minimal. Thus factors other than regionality may better explain differences in the coping strategies of parents of children with ASD.

A study investigating the lived experience of having a child in regional Canada reported parents felt isolated regarding how to best support and parent their child with ASD (Hoogsteen & Woodgate, 2013a). People living in LDP areas may lack viable formal support options, such as limited access to: adequately trained support professionals, mental health services, and information, and increased travel time to obtain necessary supports due to having to travel vast geographical distances. These factors may restrict parents' ability to adopt problem-focused and positive coping strategies and encourage the use of avoidant coping strategies (Hoogsteen & Woodgate, 2013a; Lai et al., 2015; McAuliffe et al., 2017; McAuliffe et al., 2016). Lastly, coping and emotional regulation are context bound; hence, avoidant coping strategies may be culturally normative and valued differently across contexts. That is, parents from LDP areas may place a high value on avoidant coping strategies, such as distraction, and find them effective in reducing their depressive symptoms, thus explaining the higher preference for using distraction in this population (Nolen-Hoeksema, Wisco, & Lyubomirsky, 2008).

Current evidence suggests parents who use avoidant coping styles may, over time, be at higher risk of experiencing mental health difficulties, such as depression and poor emotional regulation, given the higher incidence of these mental health conditions in people who adopt maladaptive coping strategies compared to more positive and active approaches (Benson, 2010; Compas, Connor-Smith, Saltzman, Thomsen, & Wadsworth, 2001; Hastings et al., 2005; Pisula & Kossakowska, 2010). Although avoidant coping

strategies may be helpful in decreasing stress in the short term, the likelihood of experiencing depression and emotional dysregulation in parents with a child with ASD are increased if avoidant coping strategies are recruited in lieu of more positive coping strategies, such as engagement and positive reframing (Benson, 2010; Dardas & Ahmad, 2015). Therefore, practitioners working with families of children with ASD should be cognisant of the coping styles within their unique context used by parents to deal with the demands of parenting and implement evidence-based strategies accordingly, regardless of geographical location.

The small likelihood in preferring avoidance coping styles and the absence of significant differences in other coping style preferences between parents living in LDP and DP areas may indicate regionality has little association with the coping styles used by parents of children with ASD. Factors other than regionality may have a more substantial association with the preferred coping strategies for parents of children with ASD. A review by Lai and Oei (2014) reported parent gender and age, child age, the magnitude and changing nature of their child's challenging behaviours, time since diagnosis and cultural effects are the most important factors that influence the way parents with ASD cope. Notably, respondents to the survey were mostly mothers (91%); however, between-group differences were not significant for gender when comparing DP and LDP groups. Furthermore, when sensitivity analyses were conducted by including parent gender, parent's age, child gender, and time since the diagnosis was made as covariates in the model, no statistically significant associations were evident between DP and LDP areas. Future studies investigating coping strategies in parents of children with ASD living in LDP areas should aim to capture mental health issues, in addition to experiences of stress and QoL. Further investigation into the association between coping mechanisms, psychological adjustment and impact on mental health is warranted for parents of children

with ASD, regardless of geographical location. Finally, there is a need for longitudinal studies to examine the impact of raising a child with ASD on mental health, in addition to stress levels, QoL, coping mechanisms and daily routines of families living in LDP over time. Longitudinal studies will better capture the dynamic nature of raising a child with ASD as they transition through various life stages, from the time a diagnosis is made, to school entry, entering puberty, and entering the workforce.

### **3.5.2 Stress and quality of life**

Findings from this study provide further support for the notion that parents of a child with ASD experience higher levels of stress and poorer QoL than parents with typically developing children (Baker-Ericzén, Brookman-Frazee, & Stahmer, 2005; Higgins et al., 2005; Montes & Halterman, 2007). Despite parents of children with ASD experiencing higher levels of stress and lower QoL than the general population, findings from this study suggest no overall difference in the stress levels between the parents of children with ASD living in LDP areas when compared to those living in DP areas. Limited between-group differences suggest that while it can be challenging for parents of a child with ASD living in LDP areas, these challenges do not increase stress or reduce QoL when compared to parents of a child with ASD living in DP areas who may have better access to services (Farmer & Reupert, 2013).

This is a positive finding, suggesting the barriers to accessing services for parents living in LDP areas may be decreasing when compared to parents living in DP areas, although it is acknowledged that some families living in very LDP areas may still experience challenges in accessing services (Dew et al., 2013; Dew et al., 2012). There has been a proliferation of innovative intervention models for children with ASD and their parents, such as parent-mediated, telehealth-delivered and information communication technology-based

interventions to address challenges for LDP communities to access services (Antezana et al., 2017; Parsons, Cordier, Lee, Falkmer, & Vaz, 2019; Parsons et al., 2017). Furthermore, the impact of having limited services on stress and QoL could be ameliorated by other factors unique to living regionally, such as an increased sense of community and informal social supports (Hoogsteen & Woodgate, 2013a). Future research investigating why no difference was detected between families living in DP and LDP areas stress levels and QoL is warranted. There is a need to understand better the factors that mediate the stress levels and QoL for families with a child with ASD living in LDP areas.

### **3.5.3 Father's education and disruption to family routine**

The finding that fathers of children with ASD living in more DP areas tended to have achieved more advanced education compared to those living in LDP areas mirrors the broader social trends in Australia (Australian Bureau of Statistics, 2008). Therefore, it is plausible to assume the difference in father's education levels between families living in DP and LDP is representative of the broader Australian population, as opposed to a unique characteristic of families with a child with ASD living in low-densely populated areas. Finally, our findings indicate that disruptions to family routines are not associated with the geographical location of families of a child with ASD.

### **3.5.4 Limitations**

This study has several limitations. Firstly, an analysis of non-responders could not be conducted, which may cause possible sample bias as the survey was distributed widely through numerous networks. Additionally, a drop-out analysis could not be completed despite a dedicated drop-out section in the online survey, as no responses were received. This prevented any comparison between parents who completed the survey and those who chose not to. Secondly, the study sample reflects the geographical distribution of the

general population living in remote (5.4%) and very remote (1.4%) Western Australia (Australian Bureau of Statistics, 2008). To better understand the unique experience of parents of children with ASD living in remote and very remote locations, a disproportionate amount of participants from these areas would be needed to have been recruited into the study. While the researchers made targeted effort to recruit more participants from remote areas, the numbers were small. As a result, the study may not fully capture the experience of families living in remote and very remote regions of Western Australia. Finally, the heterogeneity of regional areas in Western Australia may not be adequately expressed in the study. While all effort was made to distribute the survey to all the regions of Western Australia, particular regions were over-represented. Therefore, care should be taken in generalising the findings to all regional and remote areas in Australia.

### **3.6 Acknowledgements**

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## Chapter 4      Randomised Controlled Trial of the Therapy Outcomes By You application

Chapter 4 reported on a study that evaluates the effectiveness of the Therapy Outcomes By You application for families living in regional Western Australia through a randomised controlled trial. The research objective was to conduct a randomised controlled trial of the Therapy Outcomes By You application as an early intervention to augment existing therapies to improve visual motor, imitation, language and social skills in children with autism spectrum disorder, aged between two and six years who reside in a regional area. This chapter contains an accepted manuscript of an article published in the Journal of Autism and Developmental Disorders, which is available online:

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The spelling and wording contained within this chapter are that of the published manuscript.



# Journal Manuscript 3

## **Title**

A randomised controlled trial of an information communication technology delivered intervention for children with Autism Spectrum Disorder living in regional Australia.

## **Authors**

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#### **4.1 Abstract**

This exploratory RCT tested the effectiveness of a tablet-based information communication technology early intervention application to augment existing therapy with the aim of improving visual motor, imitation, language and social skills in young children with ASD who reside in regional areas. Fifty-nine participants were recruited and randomised to either a therapy-as-usual group or intervention group. With the exception of the expressive language subscale on the Mullen Scales of Early Learning, no significant between-group differences were recorded for visual motor, imitation, receptive language and social skills of participants between baseline and post-intervention. When all participants were pooled and measured over time, improvements were shown in receptive and pragmatic language and social skills; these gains were maintained, thus suggesting skill acquisition.

**Keywords:** information technology, RCT design, parent training, early intervention



## 4.2 Introduction

Access to and availability of therapy services for children with Autism Spectrum Disorder (ASD) in regional communities is sparse (Hoogsteen & Woodgate, 2013; Hutton & Caron, 2005; Smith, Humphreys, & Wilson, 2008). The challenges in accessing the recommended 15 – 25 hours per week of quality intervention are magnified in regional communities in Western Australia (WA), which has a geographical area of 2.53 million km<sup>2</sup> (Australian Bureau of Statistics, 2017; Smith et al., 2008; Struber, 2004). Families attempting to access appropriate health and therapy services in regional Australia may encounter a number of barriers including: 1) lower levels of autism screening and delayed diagnosis; 2) widely dispersed services requiring parents to travel vast distances to access them; and 3) difficulties in retaining skilled health professionals in these regional settings (Dew et al., 2012; Keane, Smith, Lincoln, & Fisher, 2011; Smith et al., 2008; Struber, 2004).

The financial burden for families with a child with ASD is significant. The additional financial median costs for families of having a child with ASD in WA is AU\$34,900 per annum (Horlin, Falkmer, Parsons, Albrecht, & Falkmer, 2014). Loss of income due to reduced working hours, accounts for 90% (AU\$29,200) of that amount, with an average of AU\$4,800 being spent on out-of-pocket expenses for treatment. The parents' capacity to work is also impacted by the distance and frequency of travel to take their child to therapy appointments. Furthermore, the strong association between severity of ASD symptoms and increased costs suggests that effective early intervention strategies may reduce the financial burden on the family over time (Horlin et al., 2014).

Both parent-delivered and telehealth interventions are emerging as viable alternatives to face-to-face, clinician-delivered interventions for families experiencing barriers when attempting to access services; however, the evidence for these methods of delivery is still

developing (Ingersoll & Berger, 2015; McConachie & Oono, 2013; Vismara, McCormick, Young, Nadhan, & Monlux, 2013). A recent systematic review conducted by Parsons, Cordier, Vaz, and Lee (2017) concluded there is preliminary evidence to support remotely delivered parent-mediated intervention training to improve parent knowledge, increase parent intervention fidelity and improve the social behaviour and communication skills for children with ASD. However, the authors noted that further investigation is required to determine if parent-mediated interventions can fully address the gap of limited access to services and be an effective alternative intervention for children with ASD for families living in non-urban locations. Stronger evidence for the effectiveness of remotely delivered parent-mediated intervention training will provide considerable benefit to families of a child with ASD living in regional areas.

Howlin, Magiati, Charman, and MacLean (2009) reported benefits of intense and sustained therapy for a range of educational and behavioural treatment strategies that target the core symptomatology of ASD. Moreover, Dawson et al. (2010) reported significant improvements in cognitive functioning, adaptive behaviour and autism symptoms using the Early Start Denver Model, an early intensive behavioural intervention (EIBI) requiring two hour sessions, twice per day, five days a week for two years delivered by trained therapists. These interventions require significant amounts of therapy input (dosage) with highly-trained clinicians to be effective and are associated with considerable financial cost (Whalen, Liden, Ingersoll, Dallaire, & Liden, 2006). Given the large amount of resources required to implement EIBI, information communication technology (ICT) delivered interventions may hold promise as an adjunct to current services, by increasing access while maintaining treatment effectiveness in regional communities. There has been a surge in the advancements made in ICT delivered interventions to enhance social, communicative and language development for children with ASD in recent years (Ploog, Scharf, Nelson, &



Brooks, 2013). ICT interventions to improve these skills in children with ASD include, but are not limited to: games, animated talking heads, video-modelling, parent training, visual feedback, multimedia approaches with voice and visuals, and virtual reality (Ploog et al., 2013). Despite the increased prevalence of these, there remains a dearth of evidence regarding their effectiveness for children with ASD (Boucenna et al., 2014; Doenyas, Şimdi, Özcan, Çataltepe, & Birkan, 2014; Falloon & Khoo, 2014; Fletcher-Watson et al., 2016; Golan & Baron-Cohen, 2006; Ramdoss et al., 2011; Tanaka et al., 2010; Whalen et al., 2010; Whitehouse et al., 2017). Efficacy studies for ICT interventions are required to better aid clinicians and families on how to best use limited resources to achieve the maximum benefit for children with ASD.

Children with ASD tend to have an affinity to ICT due to its slower pace and the asynchronous aspects of its interface, thus making its use as a treatment method ideally suited to this population (Ploog et al., 2013; Rajendran, 2013). The advent of tablet devices, such as Apple's iPad, has resulted in a surge of ICT supported therapy interventions. While ICT has a number of advantages that can support families with a child with ASD; this form of intervention also has barriers, such as difficulty in creating a design for a software application that is suitable for all children with ASD (Tseng & Do, 2010), limited financial and time resources of parents (Ingersoll & Berger, 2015) and poor generalisation from software application to "real word" situations (Ploog et al., 2013; Silver & Oakes, 2001). Additionally, ICT use may exacerbate existing problems associated with ASD such as: 1) decreasing the opportunities for interaction between the child with ASD and the person delivering the intervention, for example teachers, parents, or therapists; 2) increasing social isolation, due to a reduced opportunity to practice social interactions; and 3) increasing the likelihood of obsessive compulsive-type behaviours and fixation on ICT device, due to children with ASD's higher tendency to persevere on computer use

(Ramdoss et al., 2011). Finally, findings from recent systematic reviews of using ICT to teach communication, social and emotional recognition skills to children with ASD are inconclusive. The authors state that despite not having enough evidence to support them as a researched-based approach they remain a promising intervention approach and continued research is warranted (Ramdoss et al., 2011; Ramdoss et al., 2012).

Therapy Outcomes By You (TOBY) is an iPad application (app) and intervention tool developed by a team of Western Australian computer scientists, psychologists and speech pathologists. The TOBY app is based on EIBI intervention guidelines supporting high intensity interventions to address individual children's needs using behavioural, educational and developmental approaches with a focus on four major skill areas: 1) visual motor; 2) imitation; 3) language; and 4) social (Moore et al., 2015; Prior & Roberts, 2012; Venkatesh, Phung, Duong, Greenhill, & Adams, 2013; Whitehouse et al., 2017). The TOBY app is not designed to replace one-on-one therapy with clinicians, but rather to provide a low cost and easily accessible intervention method to supplement existing therapy. For families living in regional areas, this may mean decreased travel time and time off work as the frequency of face-to-face visits could be reduced, thereby reducing the economic burden for these families. The TOBY app is child specific, starting at the child's current level of functioning and progresses through the curriculum relative to the child's ability and rate of development. The TOBY app can be utilised by parents and their children with ASD without direct input from health professionals. Because the TOBY app can be completed in the homes of families under the guidance of caregivers, this novel intervention method holds significant promise for enabling regional families' access to services they may have otherwise have missed out on (Moore et al., 2015; Venkatesh et al., 2013).

A pilot study involving 33 families with a child (16 years or younger) with ASD who used the TOBY app for four to six weeks indicated that it can make a useful contribution to early intervention for children with ASD by increasing the number of learning opportunities (Moore et al., 2015). However, this study lacked both independent pre- and post- intervention measures of functioning, as well as a control group for comparison. Additionally, a recent randomised controlled trial (RCT) investigated the effectiveness of the TOBY app for families mostly living in urban areas. Participants were provided with six months of intervention with fortnightly calls from researchers. The authors concluded that the TOBY app may provide a beneficial addition to existing therapist-delivered interventions for children with ASD (Whitehouse et al., 2017). As such, the TOBY app may hold promise as a complementary therapy to existing therapies, particularly for families who may have difficulty accessing traditional face-to-face therapeutic services, such as those living in regional areas. To date, the effectiveness of the TOBY app has not been investigated for children with ASD living in regional areas.

Given the proliferation of ICT interventions for children with ASD, there is an urgent need for evidence to assist families and clinicians to navigate the plethora of options when choosing effective interventions. Additionally, the lack of methodological rigor in effectiveness studies continues to add further ambiguity for families and clinicians alike (Ramdoss et al., 2012). Evidence-grounded interventions that have established treatment effectiveness and evidence of maintenance are crucial in informing families and clinicians in decision making on intervention options, particularly for families where access to therapy services may be restricted, due to vast travelling distances (Parsons et al., 2017). Finally, insights learnt by conducting research with families of children with ASD living in regional areas will inform larger-scale efficacy studies with this population from a feasibility perspective (Bowen et al., 2009).

### **4.2.1 Objectives**

The objectives of the study were to conduct an exploratory effectiveness study of the TOBY app as an early intervention to augment existing therapies to improve visual motor, imitation, language and social skills in children with ASD, aged between two to six years who reside in a regional area. Three hypotheses were proposed:

1. The change in visual motor, imitation, language and social skills of children with ASD who received the TOBY app for three months will be significantly greater than changes in a waitlisted control group.
2. The overall visual motor, imitation, language and social skills of children with ASD who received the TOBY app for three months will improve significantly from baseline; and
3. The overall visual motor, imitation, language and social skills of children with ASD who received the TOBY app for three months will maintain these skills after three months cessation of intervention.

## **4.3 Methods**

### **4.3.1 Trial design**

This was a single-site trial using a waitlisted parallel RCT design. Participants were assigned to either an immediate intervention group or a waitlisted therapy-as-usual group using computer generated sets of random allocations finalised before the start of the study. Prior to the start of recruitment, the group allocation for participants was sealed in sequentially-numbered opaque envelopes. Once the participant had given informed consent to be included in the trial, they were block randomised in pairs, with a one-to-one allocation ratio to intervention group and the waitlist group. Group allocation was conducted prior to the collection of baseline data. Researchers anticipated a study of this

kind required extended recruitment time, thus the randomisation method was adopted to ensure equal distribution of members in the comparison groups if data collection had to be ceased at any time point. All participants received a baseline assessment at the start of the data collection. In addition to receiving therapy-as-usual, the intervention group were instructed to practise at least 20 minutes on the TOBY app daily for three-months using an iPad. Participants were then re-assessed at three and six months after the baseline assessment to establish post-intervention and follow-up measurements, respectively. The waitlisted group received an iPad without the TOBY app installed and therapy-as-usual after the baseline assessment. After the waiting period of three-months, the control group received the TOBY app for three-months. The waitlisted group were then assessed at six and nine-months to establish the post-intervention and follow-up measurements. The Consolidated Standards of Reporting (CONSORT) 2010 guidelines for evidence-based reporting of RCTs were followed to report this trial (Schulz, Altman, & Moher, 2010). This RCT was registered with the Australian New Zealand Clinical Trials Registry (ACTRN12614000745640). Ethics approval was granted by the Curtin University Human Research Ethics Committee (approval number: HR123/2014) in Western Australia.

#### **4.3.2 Participants**

Fifty-nine children and their families participated in the study. Children with a developmental age between two and six years, residing in areas outside of major cities as defined by the Australian Standard Geographical Classification System (Australian Bureau of Statistics, 2011a); who had been diagnosed with ASD as determined by the Diagnosis and Statistical Manual of Mental Disorders (DSM-5) were eligible to participate in the study (American Psychiatric Association, 2013). The age range of participants was chosen given the intervention's focus on early intervention with this population (Venkatesh et al., 2013). Participants were recruited through key ASD service providers including

paediatricians, allied health clinicians, private health service providers, parent support groups, community forums, media advertisements and snowballing techniques from the southwest region of Western Australia. Participants were screened by researchers using a pre-determined protocol prior to group assignment. All potential participants were screened over the telephone. The interview contained questions pertaining to the child's age, diagnosis and medical information. If the child was deemed eligible, a face-to-face meeting was arranged. Children were included if they also had conditions commonly occurring with ASD (e.g., anxiety, attention deficit hyperactivity disorder) provided ASD was their primary diagnosis and the co-morbid condition did not significantly interfere the participant to take part in 20 minutes of therapy per day with the TOBY app.

Children were excluded from the study if they had a physical disability that prevented them from engaging in 20 minutes of therapy per day with the TOBY app. Non-idiopathic cases of ASD including genetic disorders, such as Rett's syndrome and Fragile X syndrome, were excluded (Luyster, Kadlec, Carter, & Tager-Flusberg, 2008). Children who already had been exposed to the TOBY app prior to recruitment were also excluded from the study. Once included in the trial and randomised, the intervention group were provided with an initial training session involving information and demonstration of the intervention by the researchers. At least one parent or guardian was involved in the intervention; where possible both parents/guardians were included. Informed consent was obtained prior to their participation in the study. All assessments across all time points were completed in the participant's home and face-to-face training was provided on how to navigate and use the TOBY app installed on the iPad. Participants assigned to the waitlisted group received an iPad without the intervention installed for the children to use at their leisure.

### **4.3.3 Intervention**

The Therapeutic Outcome By You (TOBY) application is delivered using a tablet device and can be accessed via the Apple iTunes® store. The TOBY app syllabus is divided into four major skills areas: 1) visual motor, which targets perception and discrimination of sensory cues, such as colour, shape, same-ness and difference; 2) imitation, which includes copying an action, design, or pre-speech sounds; 3) language, which focuses on recognition and production of object names; and 4) social, which targets inter-personal skills, such as joint attention (Venkatesh et al., 2013). The intervention was designed to augment early intervention for children between two and eight years of age. The TOBY app is delivered within a rigorous learning framework of stimulus adjustment relative to performance, with appropriate prompting and mastery criteria. The TOBY app presents a choice of tasks, drawn from a curriculum tree, based on how the child has progressed with pre-requisite tasks. Participants automatically progress through the curriculum based on skill attainment, with activities of increasing complexity and difficulty scaffolded to promote skill development. The TOBY app uses principles from Applied Behaviour Analysis (ABA) to facilitate skill acquisition. ABA is the scientific study of behaviour and its applications to socially important problems in the natural environment (Heitzman-Powell, Buzhardt, Rusinko, & Miller, 2014). Furthermore, ABA is a method of identifying problems to reduce, skills to improve, and techniques to alter the environment to support and maintain the changes (Baer, Wolf, & Risley, 1968; Heitzman-Powell et al., 2014). Once downloaded, the application includes clear instructions to parents on how to implement the entire intervention with their child. Participants in this study received a one-hour training session from the researchers (psychologists and occupational therapists) on how to navigate and use the intervention using recommendations from a pilot trial of the intervention (Moore et al., 2015).

The intervention has three methods for the delivery of therapy: solo, partner, and Natural Environment Training (NET) (Venkatesh et al., 2013). The syllabus includes a variety of activities that utilise different methods of delivery to address the four targeted skill areas. The TOBY app solo activities involved the child interacting directly with the iPad. The TOBY app partner activities required the caregiver to be an active agent in the intervention, with the application providing instructions to the caregiver to complete various activities with their child. Caregivers then inputted the result directly into the TOBY app to track their progress. The NET activities of the TOBY app aimed to generalise learning from the solo and partner activities into natural situations by educating, prompting, and logging the caregiver's translational intervention with their child. The TOBY app was designed to address the poor generalisation of skills learned in most ICT delivered interventions to natural environment (Silver & Oakes, 2001). The NET activities also provided learning opportunities for children with ASD to apply the learned skills in the 'real world' (Hopkins et al., 2011; Ploog et al., 2013).

Participants were instructed to use the application for 20 minutes once-per-day at a time convenient to the parent and child, based on the findings of a pilot trial on effectiveness of the TOBY app (Moore et al., 2015). Researchers only followed-up with participants during the intervention period if they were contacted regarding technical issues. This level of support differs from the study by Whitehouse et al. (2017) whereby participants received a phone call from researchers to follow-up every two weeks throughout the intervention period. The decision to provide limited support was made to simulate the real-world experiences of families living in regional areas and the ability for families to download the intervention from Apple iTunes® without therapist input with the aim of gaining insight from a feasibility point of view.



#### **4.3.4 Demographic information**

Demographic information was collected from all participants. The Socio-Economic Index For Areas (SEIFA) deciles based on postcode were used to determine the socioeconomic status of the participants (Australian Bureau of Statistics, 2011b). The SEIFA is developed by the Australian Bureau of Statistics to rank areas according to their relative socio-economic advantage and disadvantage based on information from a five-yearly Census (Australian Bureau of Statistics, 2011b).

A series of standardised repeated measures were administered at baseline (T1), post-test (T2) and at follow-up (T3) to assess the effectiveness of the TOBY app. These measures included: 1) Mullen Scales of Early Learning (Mullen, 1995); 2) Symbolic Play Test (Lowe & Costello, 1988); and 3) Communication and Symbolic Behavior Scales (Wetherby & Prizant, 2002). Additionally, a 20-minute video of spontaneous interaction with a neurotypical person was obtained at the baseline (T1), post-test (T2) and at follow-up (T3). The video footage was analysed by two experienced allied health clinicians who were blinded to all aspects of the study using two standardised observation measures, the Test of Playfulness (ToP) and the Pragmatic Observation Measure (POM) (Bundy, 2004; Cordier, Munro, Wilkes-Gillan, Speyer, & Pearce, 2014).

#### **4.3.5 Primary outcome measures**

##### **4.3.5.1 Mullen Scales of Early Learning (MSEL)**

The MSEL is a comprehensive measure of cognitive functioning for infants and preschool children, from birth to 68 months. The measure consists of five scales that assess a child's abilities in visual reception, gross motor, fine motor, receptive language and expressive language (Mullen, 1995). For the purposes of this study, the gross motor scale was not administered as it was not age appropriate. The MSEL is deemed to be appropriate and

valid in assessing children with ASD (Akshoomoff, 2006). A number of participants received T-Scores at or below 20, thus representing 3 or more standard deviations below the mean. Subsequently, the decision was made to calculate developmental quotients (developmental age divided by chronological age, multiplied by 100) for the four scales used. Reliability estimates are moderate (range 0.75-0.83) with a composite median value of 0.91. Inter-rater reliability was very high, with a range between 0.91 to 0.99 (Mullen, 1995).

#### **4.3.5.2 Communication and Symbolic Behavior Scales Developmental Profile Caregiver Questionnaire (CSBS)**

The CSBS is a parent-reported standardised assessment tool designed to examine communicative, social affective, and symbolic abilities of children (Levy et al., 2003). The caregiver questionnaire was used in this study. The CSBS has strong test re-test reliability ( $r = 0.64-0.91$ ) for the checklist questionnaire and behaviour sample. The measure has moderate to strong concurrent validity ( $r = 0.59-0.61$  and  $0.65-.071$ ) with reference to the one-page parent report checklist and behaviour sample (face-to-face) of the same assessment, indicating it is a valid tool for evaluating the communication and language skills of young children (Wetherby, Allen, Cleary, Kublin, & Goldstein, 2002). The scales have been found to exhibit high internal consistency (Cronbach's alpha range 0.86 - 0.92) (Wetherby & Prizant, 2002).

#### **4.3.6 Secondary outcome measures**

The Test of Playfulness, Pragmatic Observation Measure and Symbolic Play Test were selected as secondary outcomes measures, as hypothesised skill acquisition resulting from the TOBY app use could be developmental precursors to play, pragmatic language and symbolic play skills.

#### **4.3.6.1 Test of Playfulness (ToP)**

The ToP is a 29-item observer rated instrument suitable for children between 6 months and 18 years. The ToP measures the concept of playfulness as a reflection of the combined presence of four elements: internal control, freedom from unnecessary constraints of reality, intrinsic motivation, and ability to give and read social cues (Bundy, 2004). The ToP has evidence for excellent inter-rater reliability, supported by data from 96% of raters who fitted the expectations of the Rasch model. Based on raw scores, Bundy (2004) reported moderate test–retest reliability with significant intraclass correlation 0.67 ( $p < 0.01$ ). The measure has good construct validity, data from 96% items and 98% of people fit Rasch expectations (Bundy, 2004).

#### **4.3.6.2 Pragmatic Observation Measure (POM)**

The POM is a 27-item observer rated instrument of pragmatic aspects of language based on direct observation (Cordier et al., 2014). The POM has evidence for excellent internal consistency (Cronbach's  $\alpha > 0.98$ ) and inter-rater reliability (0.89). It has good construct validity (0.55-0.77), excellent criterion validity (0.95) and high responsiveness to change (sensitivity = 79.7%; specificity = 89.6%).

#### **4.3.6.3 Symbolic Play Test (SPT)**

The SPT evaluates children's spontaneous non-verbal play activities in a structured situation. It is a simple, quick and easy to administer assessment that consists of four independent situations. Minimal prompting is provided by the assessor throughout the assessment. The assessment is independent of verbal comprehensions and expression on the child's part (Lowe & Costello, 1988; Stanley & Konstantareas, 2007). The measure has good internal consistency (Cronbach's  $\alpha = 0.81$ ) and test-retest reliability (0.64-0.81).

#### **4.3.7 Sample size**

Sample size was based on a Cohen's  $d$  of 0.8, given an  $\alpha$ -value of 0.05 and a power of 80%. Using these parameters, 24 children with ASD in each group ( $N=48$ ) were required to generate sufficient power of the study based on the effect sizes of similar studies investigating the effectiveness of ICT delivered interventions (Ramdoss et al., 2012). To account for attrition, the aim was to recruit 60 children into the study. A total of 60 families were recruited for the study, with 59 children completing baseline measurements and one withdrawing after randomisation, but before baseline assessments. Seventeen families withdrew during the course of the study; available data were included for analysis.

#### **4.3.8 Statistical methods**

Data were cleaned and analysed using SPSS<sup>®</sup> 24 (IBM Corp., 2016). Descriptive statistics were used to describe the sample. Independent  $t$ -tests at group level for continuous data and Pearson's  $\chi^2$  tests for categorical data were used to compare demographic and outcome measure differences between groups at baseline. To address hypothesis one, an intention-to-treat analysis approach was used to compare the change scores from baseline to three-month data point ( $T2 - T1$ ). Multiple imputation was used to manage the missing data. Multiple imputation is a flexible approach to missing data and is appropriate for use in RCTs (Sterne et al., 2009). There was approximately 15% missing data. Sensitivity analyses were conducted post-hoc to compare findings using the Last-Observation-Carried Forward (LOCF) and list-wise deletion methods. These conservative approaches to sensitivity analysis were chosen to ensure the impact, effect and influence of assumptions did not change the overall conclusions of the study (Thabane et al., 2013).

The effectiveness of the intervention was analysed by measuring the change scores between baseline ( $T1$ ) and three-month ( $T2$ ) data points. A positive increase in the value

indicated a positive change. Shapiro-Wilks tests were conducted to test for normality. Independent *t*-test and Mann-Whitney *U* for group comparison using the randomisation allocation as grouping variable were conducted. A *p*-value of <0.05 indicating statistically significant result was adopted with all statistical tests. Cohen's *d* scores were used to calculate effect size, where the magnitude effect sizes was interpreted as small  $\geq 0.20$ ; medium  $\geq 0.50$ ; or large  $\geq 0.80$  (Cohen, 1992). As this is an exploratory study aimed at generating hypotheses for future larger scaled studies and with consideration to not increase the likelihood of equally important Type 2 errors, we did not apply Bonferroni (or other similar) adjustments for Type 1 errors (Armstrong, 2014; Perneger, 1998).

To address hypotheses two and three, data from the intervention ( $n=30$ ) and waitlisted group ( $n=27$ ) were combined ( $n=57$ ) to analyse the changes in outcomes measures at baseline measure (T1), post-intervention (T2) and follow-up (T3) using the fixed effect of time. Linear mixed models with post-hoc pairwise comparisons were used to compare, firstly T1 to T2; secondly T2 to T3; and finally, T1 to T3. A linear mixed model was used given its suitability for modelling changes over time and the ability to accommodate missing values (Krueger and Tian, 2004). In the model, an autoregressive covariance matrix to define within-subject error was chosen with coefficients estimated via maximum likelihood. Finally, dosage was assessed as a covariate in the linear mixed models.

## **4.4 Results**

### **4.4.1 Participant flow**

Ninety-seven families and participants expressed interest in taking part in the study. Thirty-seven participants were excluded due to: a) not meeting the inclusion criteria ( $n=10$ ); b) declining to participate after receiving more information about the trial ( $n=13$ ); and c) researchers did not receive any further contact after the initial expression of interest ( $n=14$ ).

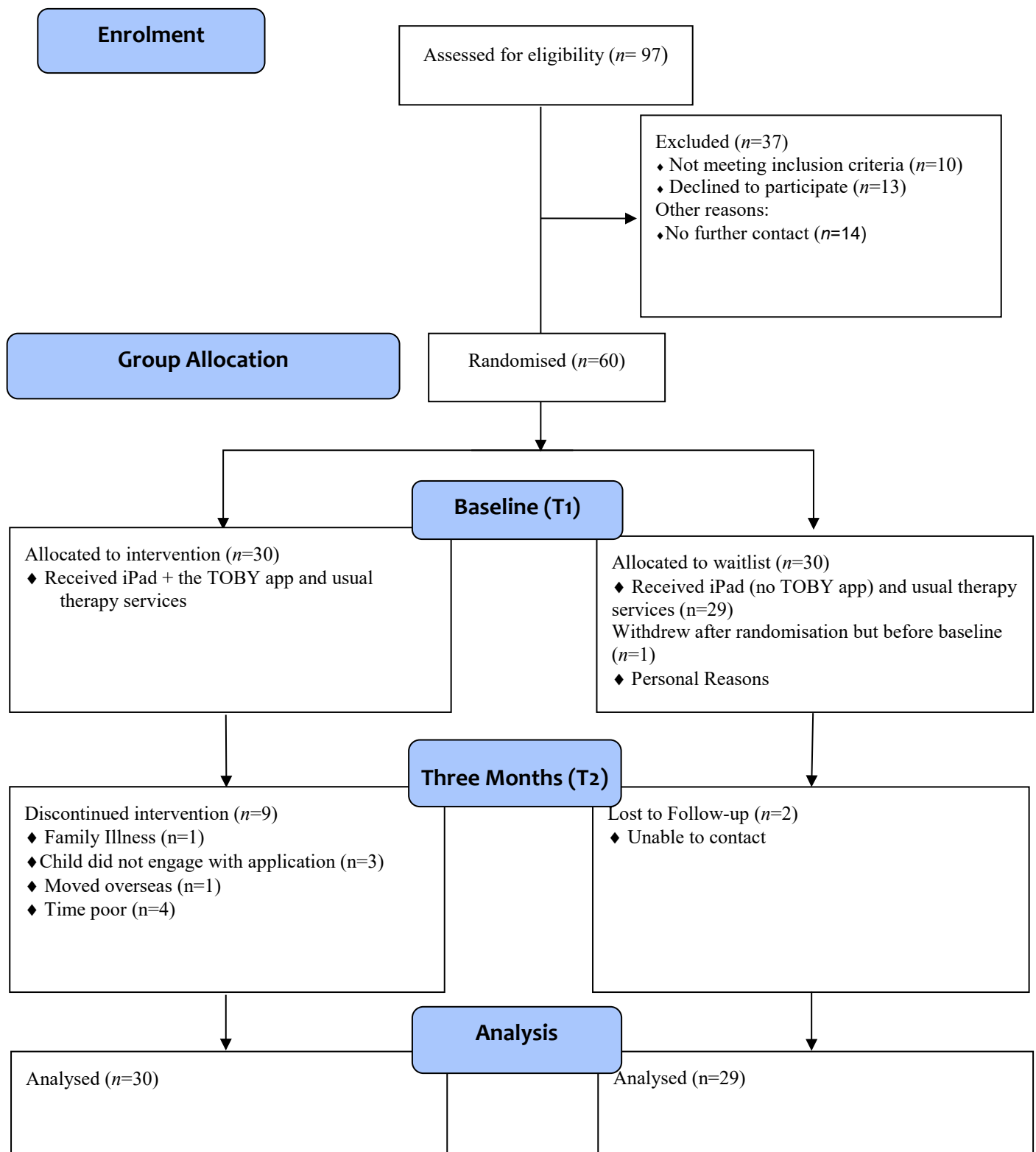
The remaining 60 participants were then randomised into the intervention ( $n=30$ ) and waitlist groups ( $n=30$ ). One participant withdrew after the randomisation process but before the baseline assessment leaving 59 participants recruited into the study. Participant enrolment is schematically presented in the CONSORT flow diagram in Figure 4.1.

#### **4.4.2 Baseline data**

No differences between groups were detected for age, gender, number of children with ASD, maternal and paternal education, population density of area or socioeconomic status ( $p > 0.05$ ). Demographic baseline data are summarised in Table 4.1. Furthermore, no differences were observed between groups in baseline data for all outcome measures ( $p > 0.05$ ).

#### **4.4.3 Dosage and feasibility**

The treatment time of participants with the TOBY app was measured by the minutes the participants engaged the device and the individual items attempted on the application per day. The activities of iPad were recorded individually on a remote server, which were retrieved for analysis after the intervention period. Participants in the intervention group on average used the application for 11.3 minutes per day (range 0 to 50.44, SD = 16.2), just over half of the recommended 20 minutes per day (Moore et al., 2015). The mean number of items attempted by the intervention group were 1,133 (range 2 to 3,017, SD = 874).



**Figure 4.1 CONSORT flow diagram showing study enrolment analysis for hypothesis one**

**Table 4.1 Child and family characteristics by group**

		Intervention ( <i>n</i> =30)	Waitlist ( <i>n</i> =29)	<i>p</i> value
Age in months	Mean (SD)	64.4 (22.73)	60.8 (15.47)	0.478
Gender	Male	25 (83.3%)	23 (79.3%)	0.692
	Female	5 (16.7%)	6 (20.7%)	
Number of Children with ASD	1	27 (90.0%)	24 (82.7%)	0.413
	2 or more	3 (10.0%)	5 (17.3%)	
Maternal Education	Diploma or Below	23 (76.6%)	23 (79.3%)	0.897
	Bachelor Degree or above	7 (24.4%)	6 (20.7%)	
Paternal Education	Diploma or Below	23 (76.6%)	24 (82.7%)	0.598
	Bachelor Degree or above	74 (24.4%)	5 (17.3%)	
Population density of area*	Inner Regional	20 (47.6%)	22 (52.4%)	0.515
	Outer Regional	9 (56.3%)	7 (43.8%)	
	Remote	1 (3.3%)	0 (0.0%)	
SEIFA Decile	Mean (SD)	5.5 (1.76)	6.0 (1.65)	0.297

*Note:*\*Based on the Australian standard geographical classification system (Australian Bureau of Statistics, 2011a). The categories include major cities, inner regional, outer regional, remote, and very remote based on a number of variables including population size and distance by road to service centers.  
SEIFA: Socio-economic index for areas



#### **4.4.4 Hypothesis one: Difference in change between intervention and waitlisted groups**

The analysis of data at three months after the commencement of trial showed that there was no between-group difference in the change of mean score (T2-T1) on the SPT, CSBS, POM and ToP ( $p > 0.05$ ) in the intervention and waitlist group (see Table 4.2). With the exception of the expressive language subscale on the MSEL, that demonstrated medium effect size (Cohen's  $d = 0.57$ ), no between-group differences in other subscales of the assessment were recorded. When compared with the waitlisted group, eight out of the 10 outcome measures in the intervention group showed greater change in mean scores; however, they were not statistically significant. Consequently, there were no evidence to support the hypothesis that there was an improvement in the TOBY app mean scores in visual motor, imitation, receptive language, and social skills of children with ASD who have used the TOBY app for three months compared to the change in visual motor, imitation, receptive language and social skills of the waitlisted group who received therapy-as-usual. Results for hypothesis one should, however, be interpreted with caution, given multiple comparisons and the relatively small group sizes ( $n=21$  for intervention group and  $n=27$  for control group).

#### **4.4.5 Hypothesis two: Overall changes in skills**

The pre-post intervention analysis of the pooled participant data ( $n=57$ ) showed changes in the sub-scale of receptive language in the MSEL, social and symbolic subdomains of the CSBS, and the POM (see Table 4.3). A trend of improvement in mean scores in all outcome measures was observed between the two time-points (T1 and T2).

Post-hoc analysis using pairwise comparisons demonstrated a significant improvement from T1 to T2 for the receptive language subdomain of the MSEL, but no significant difference between T1 and T3. Moreover, statistically significant differences between T1

and T2, as well as between T1 and T3 were observed in the social and symbolic subdomains of the CSBS. The POM demonstrated statistically significant differences from T1 to T2 and from T1 to T3. Finally, interaction effects analyses between the amount of dosage received and the MSEL, social and symbolic subdomains of the CSBS, and the POM yielded no significant results; however, the CSBS social and symbolic subdomains, and POM were approaching significance, indicating a weak moderator effect. Overall, this indicates that skills of receptive language and social skills significantly improved from pre-intervention to post-intervention for all children. Notably, the mean improvement was greater than the time elapsed, indicating the change was not just typical development. Therefore, the results partially supported hypothesis two.

#### **4.4.6 Hypothesis three: Skills learning using the TOBY app will be maintained at the three month follow-up assessment**

Three months after the TOBY app intervention period ceased, the MSEL receptive language subscale, CSBS social and symbolic subdomain and the POM remained unchanged. Most of the other scores showed a positive trend after the intervention stopped, with the exception of the MSEL subscales (see Table 4.3). The findings show that the receptive and pragmatic language and social skills of the participants were maintained after the intervention ceased for three months. Therefore, the results partially support hypothesis three.

**Table 4.2 Means scores at baseline and three months by group**

	Baseline (T1)		Three months (T2)		Change in mean score (T2 – T1 (SD))		Group	
	Intervention (n=30)	Waitlist (n=29)	Intervention (n=21)	Waitlist (n=27)	Intervention (n=21)	Waitlist (n=27)	t or z	difference p-value
MSEL	64.8 (26.27)	74.71 (26.26)	73.0 (32.13)	78.0 (23.42)	7.5 (25.4)	3.1 (20.70)	1.018	0.309
Fine Motor	60.6 (20.40)	62.9 (23.41)	68.8 (21.74)	70.2 (19.42)	7.2 (15.15)	1.4 (13.99)	0.209	0.835
Receptive Language	61.8 (26.00)	64.9 (23.75)	72.1 (32.84)	71.6 (26.98)	10.7 (14.56)	7.2 (20.93)	0.581	0.561
Expressive Language	56.5 (22.03)	63.1 (22.28)	70.9 (27.97)	67.5 (22.75)	12.3 (18.79)	3.5 (10.78)	2.136	0.033*
SPT	29.6 (7.30)	28.9 (7.72)	31.7 (6.22)	30.9 (6.85)	1.8 (5.24)	2.9 (4.29)	0.005	0.987
CSBS	33.7 (6.97)	28.1 (9.20)	36.2 (7.07)	31.0 (8.40)	3.4 (4.97)	3.2 (4.50)	-0.858	0.391
Speech Domain <sup>§</sup>	32.5 (9.66)	30.8 (8.61)	32.3 (10.67)	32.4 (10.76)	1.4 (3.58)	2.4 (15.48)	-4.32	0.661
Symbolic Domain	39.7 (8.61)	35.2 (12.09)	39.4 (11.82)	37.1 (12.08)	4.1 (2.91)	2.3 (8.80)	-1.01	0.311
POM	19.3 (40.86)	28.5 (41.79)	30.2 (31.6)	28.7 (36.77)	14.6 (42.12)	-1.7 (34.44)	1.013	0.311
TOP	50.5 (14.28)	54.0 (19.53)	54.9 (16.81)	57.3 (16.85)	4.9 (16.46)	2.4 (18.32)	0.438	0.661

\*Significant difference between intervention and waitlist group

<sup>§</sup>Ceiling Effect (> 15% participants achieved ceiling scores)

MSEL: Mullen Scales of Early Learning

SPT: Symbolic Play Test

CSBS: Communication and Symbolic Behaviour Scale

POM: Pragmatic Observation Measure

TOP: Test of Playfulness

**Table 4.3 Linear mixed model – means and standard error for all baseline, outcome and follow-up measures pooled**

		Baseline (T1)	3-Month (T2)	Follow-up (T3)	<i>F</i> - score	<i>p</i> value
		( <i>n</i> =57)	( <i>n</i> =44)	( <i>n</i> =42)		
MSEL	Visual Reception <sup>§</sup>	70.1 (3.99)	75.0 (4.34)	72.9 (4.39)	0.949	0.391
	Fine Motor	65.0 (3.19)	70.3 (3.52)	64.9 (3.6)	1.971	0.147
	Receptive Language	65.5 (4.00)	72.7 (4.26)	67.8 (4.40)	3.461	0.039*
	Expressive Language	61.1 (3.68)	67.0 (4.17)	63.9 (4.00)	1.229	0.298
SPT	Age Equivalent <sup>§</sup>	30.3 (0.98)	30.3 (1.05)	30.6 (1.05)	0.049	0.952
CSBS	Social Domain	31.9 (1.16)	34.3 (1.18)	34.7 (1.18)	13.226	<0.001*
	Speech Domain <sup>§</sup>	31.5 (1.49)	33.1 (1.52)	33.5 (1.52)	2.851	0.066
	Symbolic Domain	37.1 (1.65)	39.7 (1.67)	40.7 (1.69)	7.675	0.001*
POM		22.9 (5.17)	33.2 (5.65)	38.4 (5.73)	4.458	0.015*
TOP		53.4 (2.32)	55.7 (2.60)	58.6 (2.60)	2.185	0.119

\*Significant difference between intervention and waitlist group

<sup>§</sup>Ceiling Effect (>15% participants achieved ceiling scores)

MSEL: Mullen Scales of Early Learning

SPT: Symbolic Play Test

CSBS: Communication and Symbolic Behaviour Scale

POM: Pragmatic Observation Measure

TOP: Test of Playfulness

## 4.5 Discussion

In this study we conducted an exploratory RCT of an ICT intervention (the TOBY app), targeting the domains of motor, imitation, language and social skills of children with ASD living in regional Australia. To the authors' knowledge, this is one of very few studies to adopt a RCT design to test the effectiveness of an ICT intervention with this population. For hypothesis one, the expressive language subscale of the MSEL was the only statistically significant difference between the intervention and waitlisted groups between baseline and post intervention. For hypothesis two and three, when all the participants'

scores were pooled and measured over time, statistically significant improvements were shown in receptive and pragmatic language and social skills and these gains were maintained, thus suggesting skill acquisition. These findings indicate limited effectiveness of the TOBY app for families living in regional areas. However, this was an exploratory study with a lower intervention dosage and fidelity than prescribed and a high participant drop-out rate. Therefore, the results need to be interpreted with due caution.

The findings are largely consistent with the inconclusive evidence supporting ICT delivered interventions to improve children with ASD's social and language skills, including a study investigating the same intervention (Fletcher-Watson et al., 2016; Ramdoss et al., 2012; Whitehouse et al., 2017). Interestingly, the Whitehouse et al. (2017) study did not report improvement in expressive language, even though they used the same intervention. For the remainder of the matched outcome measures across the two trials, the Whitehouse et al. (2017) study reported improvements in the Visual Receptive and Fine Motor subscales on the MSEL, whereas no improvements for these scales were reported in this study. The main differences between the two trials were: 1) participants from the Whitehouse et al. (2017) study were mostly recruited from city areas, whereas all participants in this study were recruited exclusively from regional areas; 2) dosage was prescribed at six months for the Whitehouse et al. (2017) study, compared to three months in this study; 3) participants in the Whitehouse et al. (2017) study received fortnightly support via a phone call from researchers, whereas only technical support, if required, was provided to participants in this study; and 4) although not intentional, children in this study (mean age = 64.4 months, SD = 22.73) were almost twice the age of the children in the Whitehouse et al. (2017) study (mean age = 35.26 months, SD = 8.8). All other differences between the two studies were completely by chance. It is likely that the participants in each study would have had access different levels of therapy and education support given the

known disparity in these services between urban and regional locations; however, this could not be confirmed (Dew et al., 2012; Keane et al., 2011). Furthermore, differences in the participants' age may have influenced the variation in findings between the studies, as the TOBY app tasks may be more developmentally appropriate to the younger age cohort in the Whitehouse et al. 2017 study.

#### **4.5.1 Dosage and feasibility**

Dosage and fidelity of the intervention was poor, with participants in this study utilising the TOBY app for just under half of the recommended time. As previously mentioned, participants in the Whitehouse et al. (2017) study received six-months of the TOBY app compared to the three months in this study. Notably, Whitehouse et al. (2017) reported that their cohort utilised the application for an average of 19 minutes per day during the first three-months, dropping to an average of two minutes in the subsequent three-months. In comparison, participants in this trial used the TOBY app for 11 minutes per day on average. Researchers from the Whitehouse et al. (2017) study made fortnightly phone calls to their participants to provide support and encourage engagement. A similar level support was not provided in this study, which was a deliberate decision made in an attempt to replicate real-world experiences and inform future research with this population; that is, families who live in underserved areas may not have access to high levels of support and follow-up from clinicians and researchers (Hoogsteen & Woodgate, 2013). However, as a consequence of the low treatment adherence, the usage of the TOBY app may not have been intensive enough to facilitate skill gains in the areas targeted by the intervention. Conversely, the three-month intervention duration seemed the appropriate length given the high drop-off in usage in the second three months reported by Whitehouse et al. (2017); hence, the shorter intervention period was unlikely to have an impact on the overall effectiveness of the intervention. When considering all these factors collectively, the higher

levels of support provided in the Whitehouse et al. (2017) study did not appear to influence the results overtly when compared to a reduced level of support provided in this study.

#### **4.5.2 Participant retention**

Participant drop-outs in this study were notably high, with poor engagement by children being cited as one of the main reasons. When doing between-group comparisons, the drop-out rate was approximately 15% ( $n=13$ ) with missing data increasing to approximately 20% ( $n=15$ ) when analysing within-group differences over time. This suggests that low treatment adherence may have been the result of the intervention not being engaging enough for participants. For ICT intervention developers and clinicians who may recommend these modalities, it is worth noting that interventions need to incorporate more advanced skills that need to be mastered to facilitate continued use, as children may disengage if the activities are not challenging enough (Allen & Warzak, 2000).

#### **4.5.3 Implications for future research**

Future large-scale studies investigating the effectiveness of interventions aiming to overcome the barrier of distance should include families of children with ASD living in remote and very remote locations in Australia. The participants in this study lived mostly in regional Western Australia. Therefore the potential benefit for families living in remote and very remote areas who may benefit most from an intervention of this nature was not captured. Families living in remote areas experience a vastly different therapy landscape from that of city and regional areas given the poorer infrastructure, disparate population and difficulties retaining adequately trained staff, so effectiveness studies focussing specifically on this population is warranted (Dew et al., 2012; Smith et al., 2008; Struber, 2004). Findings from the Whitehouse et al. (2017) study indicated that ongoing support could improve intervention dosage and fidelity. For future research with this population,

innovative models of service delivery could incorporate the recruitment and capacity building of local therapists, educators, disability support workers, or allied health assistants to support families who are undertaking early intensive behavioural interventions (Dew et al., 2012). The increased support provided by these professionals may help to ameliorate poor intervention dosage and fidelity observed in this trial. Furthermore, other ICT strategies such as regular videoconferences by trained therapists, either directly with parents or with local therapy assistants, may offer promise in increasing engagement and support in ICT interventions for families with a child with ASD (Dew et al., 2012; Ingersoll & Berger, 2015).

Further research should investigate the appropriateness of the intervention for families with a child with ASD living in regional areas. Participant drop-out due to time restrictions and children not engaging with the application were evident. From a feasibility perspective, increased insight into these phenomena with this population may help future developers and researchers create strategies to mitigate potential factors resulting in poor engagement. In-depth interviews investigating the user experience of the intervention may also contribute to developing a deeper understanding of complex interplay between these factors. Despite the limited research evidence of their effectiveness, ICT delivered interventions for children with ASD continue to proliferate at a rapid rate. Ongoing efficacy studies with robust methodologies on ICT delivered interventions are required to help inform parents and clinicians of the most effective and appropriate intervention for children with ASD.

#### **4.6 Conclusion**

This exploratory study suggests that when compared to therapy-as-usual, the TOBY app provides benefit in expressive language for children with ASD living in regional areas of



Western Australia. Additionally, significant gains were observed in the areas of receptive and pragmatic language and social skills from the intervention, suggesting skill acquisition. From a feasibility perspective, dosage and intervention fidelity were poor, indicating a need to develop and implement evidence-based strategies aimed at improving adherence to ICT delivered interventions for children with ASD.

## 4.7 References

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## Chapter 5      Appropriateness of the Therapy Outcomes By You application

Chapter 5 describes a study evaluating the appropriateness of the Therapy Outcomes By You application for families of children with autism spectrum disorder living in regional Australia. The study provides insight into the barriers and facilitators identified by parents who used the Therapy Outcomes By You application living in regional Australia. The study also provides recommendations to better inform the development and use of other information communication technology based interventions for clinicians, researchers and developers that use parent-mediated interventions to complement existing therapy services. This chapter contains an accepted manuscript of an article published in the Journal of Autism and Developmental Disorders, which is available online:

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The spelling and wording contained within this chapter are that of the published manuscript.



# Journal Manuscript 4

Appropriateness of the TOBY application, an iPad intervention for children with Autism Spectrum Disorder: A thematic approach.

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## **5.1 Abstract**

This study aimed to explore the appropriateness of an ICT intervention, the Therapeutic Outcomes by You application (TOBY app), from the perspectives of the parents. Parental experiences of twenty-four parents of a child with ASD who had participated in a three-month trial using the TOBY app were collected using semi-structured interviews. Thematic analysis was conducted and themes were mapped against an appropriateness framework. Collectively, parents felt the TOBY app was relevant and important to them and their children's needs, while expressing partial support of the TOBY app as: a positive experience for them and their children, beneficial for them and their children, a socially and ecologically valid intervention, and an intervention that supported change and continuation in the skills learnt.

**Keywords:** Appropriateness, information technology, early intervention





## 5.2 Background

Living with a child who has an autism spectrum disorder (ASD) can be challenging for families (Gray, 1994; Rao & Beidel, 2009). Children with ASD have communication deficits including, but not limited to, difficulty in developing age-appropriate friendships and problems interpreting nonverbal gestures (American Psychiatric Association, 2013). Children with ASD also have rigid routines with heightened sensitivity to changes in their environment (Happé & Ronald, 2008). There is a growing body of evidence on the effectiveness indicating that educational and behavioural interventions support greater social, economic and community participation for children with ASD. In particular, there is substantial evidence to support the effectiveness of Early Intensive Behavioural Interventions (EIBI) for children with ASD, such as the Early Start Denver Model (Dawson et al., 2010; Howlin, Magiati, Charman, & MacLean, 2009). However, the feasibility and appropriateness of these interventions are in question, as they require significant amounts of therapy input (dosage) with highly-trained therapists coming at a considerable financial cost (Whalen, Liden, Ingersoll, Dallaire, & Liden, 2006).

Information and Communication Technology (ICT) based interventions are emerging as a viable mechanism to provide cost-effective, direct intervention to children with ASD; however, empirical support for them remains limited due to the complexities involved with the development and investigation of these delivery models (Ramdoss et al., 2012; Wainer & Ingersoll, 2011). Interventions using a technology delivery system for people with ASD, such as computers and tablets, have made considerable advances in recent times, making them more readily accessible for families (Ploog, Scharf, Nelson, & Brooks, 2013; Ramdoss et al., 2011). With decreasing costs, increasing ease of use, and children with ASD often having a high affinity for these devices, ICT-based interventions are showing great promise as a potential platform to deliver interventions to children with ASD (Ploog

et al., 2013; Tseng & Do, 2010; Whitehouse et al., 2017).

Despite the numerous advantages of ICT-based interventions, barriers associated with this type of therapy delivery method need to be considered. The use of ICT-based interventions with this population is associated with decreased social interactions with peers, parents and clinicians, the possibility of perseveration on particular items installed on ICT devices, and poor generalisation of skills learnt (Ramdoss et al., 2011). Silver and Oakes (2001) have argued that it is not whether ICT-based interventions are superior to one-on-one interventions, rather, given the constraint of resources, how do we best optimise the use of them in combination with conventional one-on-one interventions?

The Therapy Outcomes By You application (TOBY app) is one such ICT-based intervention. The TOBY app is a tablet (iOS<sup>®</sup>) delivered intervention tool developed by a team of computer scientists, psychologists and speech pathologists to provide EIBI therapy to children with ASD (Venkatesh, Phung, Duong, Greenhill, & Adams, 2013). The TOBY app uses an Applied Behaviour Analysis (ABA) framework and is based on EIBI intervention guidelines supporting high-intensity interventions to address individual children's needs using behavioural, educational, and developmental approaches (Prior & Roberts, 2012; Venkatesh et al., 2013). The TOBY app syllabus contains four major skills areas: 1) *visual motor*, which targets perception and discrimination of sensory cues, such as colour, shape, sameness and difference; 2) *imitation*, which includes copying an action, design, or pre-speech sounds; 3) *language*, which focuses on the recognition and production of object names; and 4) *social*, which targets inter-personal skills, such as joint attention (Venkatesh et al., 2013). According to its developers, the strength of the TOBY app is its focus on teaching a parent how to teach (Venkatesh et al., 2013). The TOBY app can be used by parents and their children with ASD without direct input from clinicians;

however, it is designed to complement face-to-face therapy.

Two Australian randomised controlled trials have reported on the TOBY app's effectiveness. The first by Whitehouse et al. (2017) reported improvements in visual motor skills and fine motor skills in children with ASD aged two to six years living in Australia, while a second study by Parsons, Cordier, Lee, Falkmer, and Vaz (2019) suggested the TOBY app improved receptive and pragmatic language in children with developmental ages between two and six living in regional Australia. Both studies reported issues with dosage and intervention fidelity and called for further research into the barriers of the TOBY app use. Participants in the Whitehouse et al. (2017) study completed the intervention for six months, with the average use dropping from 19 minutes over the first three months to 2 minutes in the subsequent three months. Notably, participants in this trial received fortnightly calls from researchers to provide support and encouragement. Participants in the Parsons et al. (2019) study completed the trial for three months, at an average use of 11.3 minutes per day. Minimal support was provided by the researchers in this trial to improve ecological validity; that is, reduced access to support and follow-up for families living in regional Australia.

With the need for cost-effective and evidence-based interventions, the impact of using resource-intensive ICT-based interventions from the perspective of the end-user to ensure its appropriateness with the intended client group needs to be explored (Campbell et al., 2000; Craig et al., 2008; Evans, 2003; Hammell, 2001). Appropriateness can be defined as the perceived fit, relevance, or compatibility of an intervention for a given practice setting, provider, or consumer (Proctor et al., 2011). Evaluation of the appropriateness is vital in the overall appraisal of the value in an intervention, and a qualitative approach through interviews is recognised as an appropriate method to explore the appropriateness of an

intervention due to the approach's ability to accurately capture the subjective human experience that is often excluded from experimental research (Evans, 2003; Hammell, 2001). Moreover, qualitative methods enable researchers to identify the delivery methods and characteristics of therapeutic interventions that best address the needs and priorities of the client, a central tenet in client-centred practice (Hammell, 2001). While the evaluation of effectiveness relates to whether the intervention achieves its intended outcomes, appropriateness is more concerned with psychosocial aspects of the intervention than the physiological (Evans, 2003). That is, appropriateness is concerned with the impact of the interventions from the perspectives of the recipient. Regardless of the intervention's effectiveness, if deemed unacceptable by the end-user, poor adherence and early abandonment of the intervention may occur, therefore reducing the overall value of the intervention (Evans, 2003; Solish & Perry, 2008).

Appropriateness, in the context of this study, addresses the experience of using the TOBY app from the perspectives of the parents who participated in a waitlisted parallel randomised controlled trial (RCT) study by Parsons et al. (2019). Forty-eight participants from the Parsons et al. (2019) study completed three months of the TOBY app for a prescribed 20 minutes per day using an iPad<sup>®</sup> provided by the researchers. All participants lived in regional areas of Australia and received minimal support from researchers due to limited resourcing in the project and to simulate ecological conditions likely experienced in regional Australia. A comprehensive description of the intervention can be found in Parsons et al. (2019)'s study. To evaluate the appropriateness of the TOBY app, a five dimension framework commonly applied in allied health and therapeutic interventions as described by Evans (2003) was used, namely: 1) the intervention addresses a health issue important to the participant; 2) involvement is a positive experience for participants; 3) the outcomes are perceived by participants as beneficial; 4) the components of the intervention

are ecologically valid (logistically viable in the participants' everyday context) and 5) techniques are continued to be used once the intervention has ceased (Allan, Wilkes-Gillan, Bundy, Cordier, & Volkert, 2018; Bowen et al., 2009; Cordier et al., 2016; Nastasi et al., 2000; Wilkes-Gillan, Bundy, Cordier, Lincoln, & Hancock, 2015).

To date, there has been only one paper investigating parent's experiences of using the TOBY app using thematic analysis, with none applying an established framework to investigate its appropriateness (Rogerson et al., 2018). Further, limited research has been conducted to date into the appropriateness of ICT-based interventions for children with ASD, a crucial aspect of evaluating complex psychosocial interventions (Campbell et al., 2000; Craig et al., 2008; Smith et al., 2007). Participants in the Rogerson et al. (2018) study were sampled from an effectiveness study by Whitehouse et al. (2017) — consisting of families living mostly in major cities in Australia. Participants from the Whitehouse et al. (2017) received fortnightly phone calls and completed the TOBY app for six months. In the context of evaluating the appropriateness of the intervention, the Rogerson et al. (2018) study did not apply a theoretical framework to anchor their analytic claims, a known limitation of thematic analysis (Braun & Clarke, 2006; Rogerson et al., 2018). As a result, findings did not encompass key dimensions of appropriateness, such as the perceived importance health issue the TOBY app was addressing, the perceived benefit of the TOBY app and the continuation of the skills learnt while completing the TOBY app.

Furthermore, when compared to parents in the Rogerson et al. (2018) study, participants from this study were solely from regional areas of Australia, received minimal support from researchers, and used the TOBY app for three months. Moreover, the absence of a theoretical framework in the Rogerson et al. (2018) study, and different ecological factors during the intervention phase between the Whitehouse et al. (2017) and the Parsons et al.

(2019) study's participants support further investigation into the appropriateness of using the TOBY app from the perspective of parents living in regional Australia in the Parsons et al. (2019) study.

Using robust qualitative research methodology anchored in a theoretical framework, this study aims to evaluate the appropriateness, a key dimension in the development and evaluation of complex interventions, of the TOBY app for families of children with ASD living in regional Australia (Campbell et al., 2007; Craig et al., 2008; Evans, 2003).

Further, this study aims to provide insight into the barriers and facilitators identified by parents who used the TOBY app living in regional Australia. The findings could also provide valuable insight into ICT-based interventions and better inform the development and use of other ICT-based interventions for clinicians, researchers and developers that use parent-mediated interventions to complement existing therapy services.

### **5.3 Methods**

#### **5.3.1 Research approach**

As this was an exploratory study about parents' perceptions, the study was guided by a qualitative design using the approach to thematic data analysis outlined by Braun and Clarke (2006). Thematic analysis is an accessible and flexible method that can be used to summarise key features and themes from a large body of data and offer a thorough description of the data set (Braun & Clarke, 2006). Furthermore, this type of analysis is more descriptive than interpretive, focusing more on the description of the participant's experience and less on the interpretation by the researcher (Creswell, 1998).

#### **5.3.2 Participants**

Participants were parents of a child with ASD who had participated in a three-month RCT using the TOBY app (Parsons et al., 2019). Maximum variation purposive sampling was

used to recruit parents from the RCT study participants to minimise bias. Given the relative homogeneity of the parents based on gender, age range, and non-urban context, we maximised variation within the available sample by recruiting participants from three categories: low ( $n = 8$ ), medium ( $n = 8$ ) and high ( $n = 8$ ) levels of recorded app use in the RCT. App use was measured using back-end server data that is automatically gathered from the tablet device. Participants were ranked for use on three measures: 1) time spent using the app on the device; 2) items attempted; and 3) items completed. The rationale was to obtain a rich and overarching narrative based on information related to both the enablers and the barriers in using the intervention by gaining insights from participants with varying levels recorded use of the app. That is, exploring the different experiences from a range of participants, as opposed to making explicit and descriptive between-group comparisons. Twenty-four mothers of a child with ASD from a pool of 59 families from the RCT agreed to participate in this study. Parents were included if they had delivered the TOBY app intervention to their child throughout the intervention period of the RCT and were available to complete a telephone interview. Semi-structured interviews between 20 to 45 minutes in duration were conducted to explore the experience of the TOBY app. All participants were mothers, and their demographic information is summarised in Table 5.1.

**Table 5.1 Participant demographics**

Participant variables		(n=24)
Age (years)	Mean (SD)	37.0 (5.05)
Gender	Female	24 (100%)
Family structure	Two-parent	17 (71%)
	Single parent	7 (29%)
Number of Children with ASD	1	21 (88%)
	2 or more	3 (12%)
Average number of children in care	Mean (SD)	2.7 (1.06)
Mothers Education	Diploma or Below	20 (83%)
	Bachelor degree or above	4 (27%)
Remoteness area*	Inner Regional	16 (67%)
	Outer Regional	7 (30%)
	Remote	1 (3%)
SEIFA** Decile	Mean (SD)	5.5 (1.98)
Child variables		
Age (months)	Mean (SD)	60 (18.90)
Gender	Male	18
	Female	6

*Note:*

\*Based on the Australian standard geographical classification system (Australian Bureau of Statistics, 2011). The categories include major cities, inner regional, outer regional, remote, and very remote based on a number of variables including population size and distance by road to service centres.

\*\*SEIFA: Socio-economic index for areas

### **5.3.3 Procedures**

Ethical approval was obtained from the Curtin University Human Ethics Committee before commencing the individual interviews (approval number: HR 123/2014). There were no adverse events to report, and no participants withdrew from the study. Conflicts of interest were declared at the beginning of all interviews, with parents being fully informed the interviewer had no vested interest in the app. Travelling distances were prohibitive to conduct face-to-face interviews. Hence, phone interviews were used with the aim of being more convenient for participants. Interviews were conducted at a time convenient for



participants, occurring between February and June 2017. Parents' experiences of the TOBY app were ascertained using a semi-structured interview, exploring the following areas: 1) the child's experience using the app; 2) parents' experience using the app; 3) if parents perceived the TOBY app to be effective for their child; 4) if parents perceived the TOBY app to be effective for themselves; 5) the ease of use, including the planning needed to implement the suggested dosage; 6) the level of support required to use the app effectively; 7) their intended future use of the app; and 8) suggested improvements to the app (See Supplementary File 1). A combination of open and close-ended questions were used to explore themes and clarify meaning. Close-ended questions with yes/no options were followed by probing open-ended questions to fully capture the perspective or experience.

Interviews lasted between 16 and 45 minutes in duration, and a digital voice recorder was used to record the interviews, which were subsequently transcribed verbatim by a professional transcription service.

#### **5.3.4 Data analysis**

Systematic coding and categorisation were completed using transcriptions of the in-depth interviews (Liamputtong, 2013). Thematic analysis was performed identifying trends and patterns of words used, their frequency, their relationship, and the structures of discourses of communication (Braun & Clarke, 2006). Six steps of data analysis were followed as described by Braun and Clarke (2006): 1) familiarising oneself with the data; 2) generating initial codes; 3) searching for themes; 4) reviewing themes; 5) defining and naming themes; and 6) producing the report or article. NVivo© software was employed to manage the data by categorising and summarising data that were similar.

The semi-structured interviews were based on the literature of evaluating appropriateness

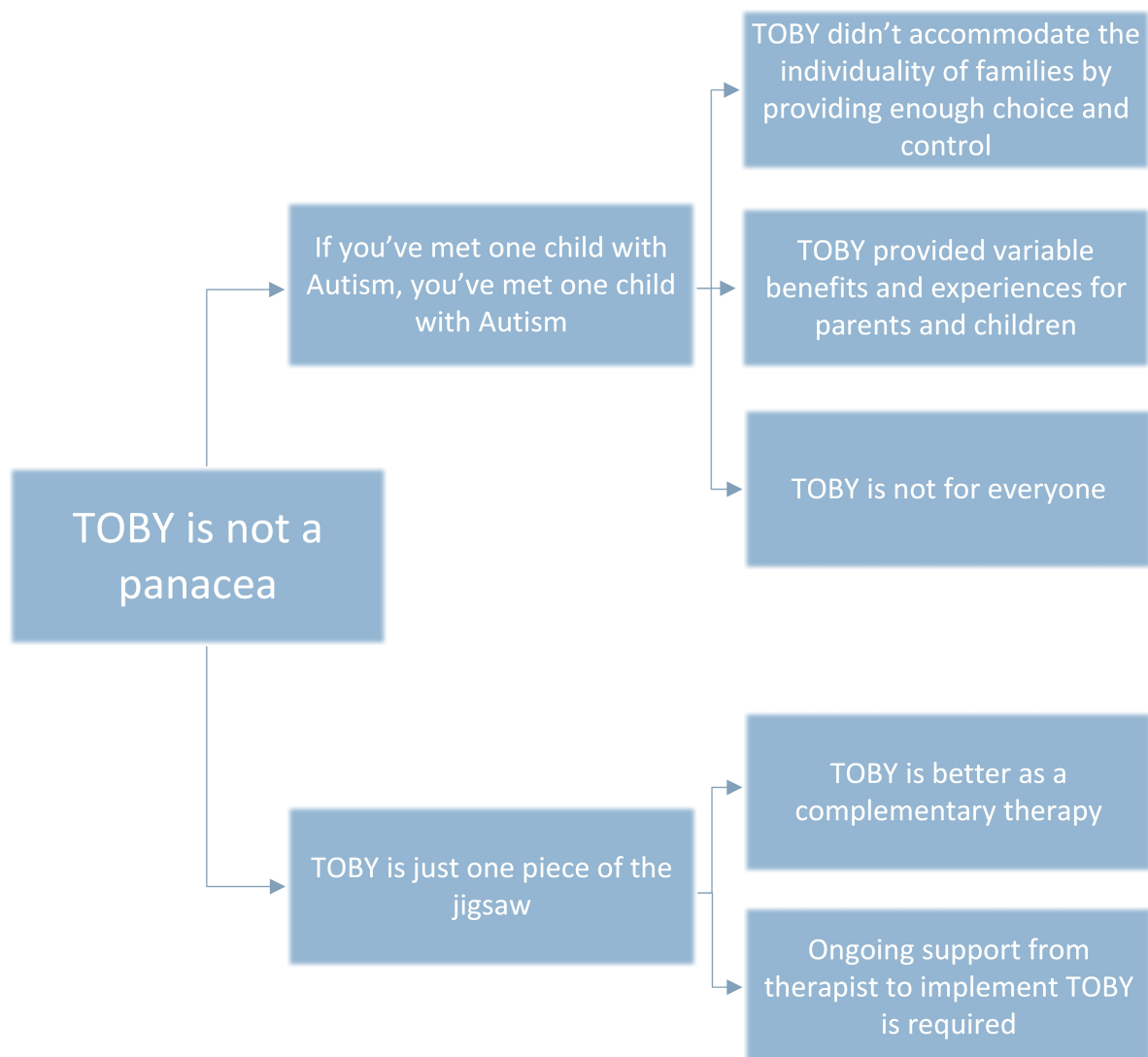
for interventions for children with ASD. That is, questions sought to explore the five dimensions of the appropriateness framework, namely: 1) the intervention addresses a health issue important to the participant; 2) involvement is a positive experience for participants; 3) the outcomes are perceived by participants as beneficial; 4) the components of the intervention are ecologically valid (logistically viable in participants' everyday context); and 5) techniques are continued to be used once the intervention has ceased (Allan et al., 2018; Bowen et al., 2009; Cordier et al., 2016; Nastasi et al., 2000; Wilkes-Gillan et al., 2015).

Trustworthiness was established based on the four strategies recommended in the literature: credibility, transferability, dependability, and confirmability (Anney, 2014; Krefting, 1991; Shenton, 2004). All interviews were conducted by the first author, an experienced occupational therapist and skilled interviewer, to enhance consistency. Throughout the data analysis, process interpretations were cross-checked over several research meetings by the second author; an experienced qualitative researcher who had no involvement in the RCT and added a non-biased and critical layer of independence to the analysis. The development of sub-themes and themes were discussed among the entire research team at several team meetings until full agreement had been reached. A clear audit trail using thematic analysis was maintained throughout the process. Finally, transcriptions were sent back to the participants for member checking, to ensure accurate recording of their responses to add further rigour to triangulation strategies. Lastly, the evaluation of the themes against the five dimensions of the appropriateness framework and their relationship strength (no support, partial support, or strong support) was discussed among all authors at several team meetings until consensus was reached. A ten-point rating scheme was also created for completion by parents to ascertain their perceptions of: 1) their skill using the technology; 2) the child's experiences of using the TOBY app; 3) their experience of using the TOBY

app; 4) benefits to the child using the TOBY app; 5) benefit to them using the TOBY app; and 6) usability of the TOBY app. These quantitative secondary data supported data collected in the interviews and triangulated themes identified (Krefting, 1991).

## 5.4 Findings

Thematic analysis of interview data led to the development of a thematic schema (see Figure 5.1). The schema visually represents the relationships between the themes. The schema consists of three levels; one core theme, two major themes, and five sub-themes. Pseudonyms for children have been used when reporting findings for confidentiality.



**Figure 5.1** Thematic schema

#### **5.4.1 Core theme: The TOBY app is not a panacea**

Overall, parents expressed that while the TOBY app was useful as a complementary intervention, it was not going to be the solution to all of their children's challenges. This led to the development of the core theme: The *TOBY app is not a panacea*. There were both data to suggest the TOBY app was effective and beneficial to both the parent and their child, as well as, data to suggest the TOBY app was not effective or beneficial for parents and their children. That is, the data did not indicate parents conclusively perceived the intervention to be beneficial for them or their children at the group level.

Parents strongly expressed the need for additional support from therapists. Parents reported the need for support in relation to challenges associated with engaging their child with the TOBY app, and the need for strategies to address problem behaviours arising from using the TOBY app. All 24 parents indicated they would recommend the intervention to a friend, suggesting while they acknowledge the TOBY app might not be beneficial for some children, they believe it holds merit and may be beneficial for other children with ASD with differing needs or interests. Moreover, this finding suggests that parents deemed the TOBY app to address issues that were relevant and important to both them and their children. When asked if they would recommend the app to their friend, one parent's comment best summarises the parent's responses: "Well, I would just tell them [about] our experience with it [the TOBY app] and that every child is different, to definitely give it a try, and if it works for them, to continue to use it."

Parents reported a broad range of the benefits of the TOBY app, as well as some challenges they experienced while implementing the intervention. This eclectic and often conflicting data lead to the development of two sub-themes (see Figure 5.1).

#### **5.4.1.1 Major theme: If you have met one child with Autism, you have met one child with Autism**

This major theme reflects the individuality of the children who received the intervention and was continually reinforced throughout the interviews. Heterogeneity of the perceived benefits, as well as the differing experience for all participants, led to the third level of the thematic schema (see Figure 1). The following comment from a parent best summarises this sub-theme: “Cos like I said, I did quite like it, just not for my Jack.”

##### **5.4.1.1.1 The TOBY app did not accommodate the individuality of families by providing enough choice and control**

Parents described the frustration of their inability to control the content which both themselves and their children were completing on the TOBY app. Parents felt the scaffolded curriculum tree was too restrictive, thus not allowing them to choose the difficulty level of the activities their children were completing. Parents reported their experience and the benefits for the children would have been improved with more choice and control about what activities their children completed on the TOBY app. The following parent comment captures this theme:

*Her receptive language is really good. So asking her to find the same or this or that or the other thing was a bit too simple for her but there was no way to 'skip' those and just go to the areas that I felt she needed I think. That was a bit of a problem for me.*

Additionally, some parents wanted more choice about the pictures of everyday items used in the app to generalise skills to real-world contexts. They felt their children would have responded better if they could take pictures of the items they had in their own house, instead of generic items included in the TOBY app.

#### **5.4.1.1.2 The TOBY app provided valuable benefits and experiences for parents and children**

Parents reported a broad range of benefits of the TOBY app; however, from the parents who suggested the TOBY app was effective, no strong consensus was evident suggesting the TOBY app was superior in the development of any particular skill. Parents reported improvements in their children across a wide variety of skill areas, such as: 1) behaviour; 2) visual-spatial skills, such as matching and visual discrimination; 3) fine motor skills; 4) daily routines, such as dressing, showering, and meal times; 5) social engagement with peers and parents; 6) joint attention; 7) cognitive tasks, such as problem-solving; 9) play; and 10) language. Additionally, parents reported the TOBY app was easy to use, with clear instructions. Some parents reported the TOBY app helped them to better understand their children's strengths and weaknesses in the skills the TOBY app was targeting, while improving their own skills in delivering EIBI to their children.

*It definitely taught me about early intervention, and it taught me what James needed and didn't need. That was important. It also gave me confidence in my perception about what he could do, and I do think it was important for the parent as well as for the child*

#### **5.4.1.1.3 The TOBY app is not for everyone**

Despite a number of parents reporting the benefits of using the TOBY app, some parents stated the app provided little benefit to their children. "I don't think there have been any huge changes over that time period. Like nothing that really 'stood out' to me at any rate." One of the main issues parents reported experiencing was difficulty in engaging their children with the TOBY app. See the comment from a parent that captures this notion:

*Paul's interest was lacking in the TOBY app. Which was a surprise 'cos looking at it with the options for fireworks and stars and all those things for the rewards that it does give and the feedback that it does give, he really had no interest.*

Parents commonly stated it was challenging to find time to complete the suggested 20 minutes per day, especially if their children were at school or attending other therapy appointments. Additionally, in the opinion of some parents, it was difficult to keep their children interested for the recommended 20 minutes per day.

#### **5.4.1.2 Major theme: The TOBY app is just one piece of the jigsaw**

This major theme captures the notion that the TOBY app is just one component of a broader therapy landscape. That is, parents were firm of the opinion the TOBY app should not be used in isolation and ongoing support, as well as other therapeutic interventions, are still required to address their children's needs. This major theme derives two sub-themes (see Figure 5.1).

##### **5.4.1.2.1 The TOBY app is better as a complementary therapy**

Most parents reported that while the TOBY app was beneficial and could demonstrate utility in decreasing the frequency of therapy sessions, particularly for families who needed to travel considerable distances to services; it could not, and should not, replace face-to-face therapy.

*Look, I think after having done it, I think that you can't replace the 'face-to-face.' So that's my opinion for my child. I also think that the App is very easy to understand and very easy to follow and we were successfully able to follow the instructions and do the tasks. I wasn't successfully able to get my child to comply. I also wasn't successfully able to figure out where we should be 'at.'*

*So you could quite easily download it from the App store and use it and follow along – there's certainly enough instructions in it. But in terms of 'best practice' for just the therapy, then I don't think that should be the sole thing that you're doing.*

Parents expressed the utility of the TOBY app for its ability to increase the amount of therapy their children receive. Parents suggested that when used in conjunction with a therapist, the TOBY app can be used to reinforce therapy goals at home by allowing the child more opportunities to practice a particular skill area.

*Yeah, it just gave him that little bit more time to work on those skills that he was trying to develop in therapy, it kind of worked like a follow-up. So you don't wait a full week to then go back and practise that skill. Even though we try and do it, it's just another backup if you like. Another way to reinforce it. The skills that he's been learning.*

Finally, parents reported that the TOBY app held potential as a tool used to receive therapy via telehealth delivery methods. Parents reported the TOBY app could assist therapists to receive objective data regarding the children's abilities in particular skill areas, which could aid in informing the therapist in their clinical reasoning from a remote location.

*It would be good for families living in a regional area that hasn't got many services. Working with TOBY [app] and in conjunction with their therapists in another area would be really good. That would be an awesome idea. At least these therapists that aren't actually face-to-face with them can actually access the app and see where they're at. So when they see the parents next, they can tell them what they may need to work on sort of thing. So it just helps everybody.*



#### **5.4.1.2.2                      Ongoing support from therapists to implement the TOBY app is required**

Parents overwhelmingly reported the need for ongoing support from therapists while completing the intervention, particularly if problem behaviours associated with the ICT use emerged or the child refused to engage with the TOBY app.

*You would probably need a therapist, just in terms of I suppose like ‘trouble-shooting’ like say when Amy couldn’t do something and she just refused to do it. Someone then who might have some ideas as to how to get around that possibly or if you feel like the child’s you know, not progressing and then what could you do or something along those lines*

*I think conferring with the therapists and seeing that part of the app where they can see where she’s at sort of thing, and they can put their input into what areas they think need to be worked on.*

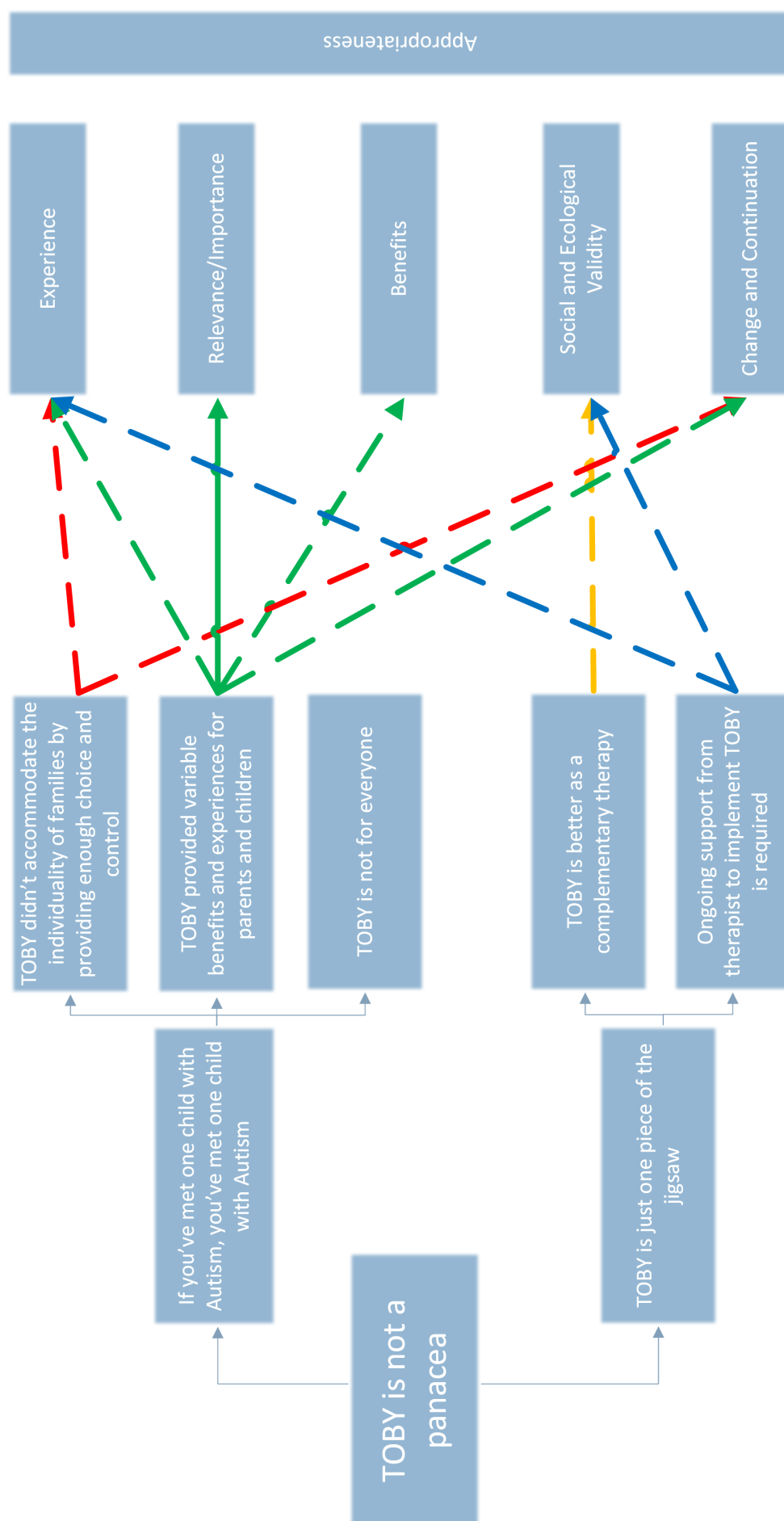
#### **5.4.2                      Evaluation of themes against an appropriate framework**

Overall, the parents’ evaluation of the TOBY app as an appropriate intervention was partially supported, when identified themes extracted from the interview were evaluated against the five dimensions of the appropriateness framework (see Figure 5.2) (Bowen et al., 2009; Evans, 2003; Nastasi et al., 2000). Broken lines in Figure 5.2 represent parents’ partial support of the intervention’s appropriateness for the corresponding dimension, whereas solid lines represent parents’ full support of intervention’s appropriateness. Collectively, parents felt the TOBY app was relevant and important to them and their children’s needs, while expressing partial support of the TOBY app as: 1) a positive experience for them and their children; 2) beneficial for them and their children; 3) a socially and ecological valid intervention; and 4) an intervention that supported change and

continuation in the skills learnt.

### **5.4.3 Results from rating scale data**

Parents generally reported positive scores on the 10-point scale for appropriateness. See Table 5.2 for a full summary of the results. The TOBY app's usability scored the highest with a mean score of 8.52 (SD 1.7). Interestingly, parents rated using the TOBY app was a better experience and more beneficial for them, compared with their children. Parents self-report of their skills in using technology was the lowest score, although this was still relatively high with a mean score of 6.91 (SD = 1.76). A one-way ANOVA was conducted for between-group comparisons for low, medium and high users and no significant differences were detected for any of the items.



**Figure 5.2 Thematic schema evaluated against an intervention appropriateness framework**

**Table 5.2 Parent ratings of quantitative questions**

<b>Question</b>	<b>Low usage<sup>a</sup></b>	<b>Medium usage<sup>a</sup></b>	<b>High usage<sup>a</sup></b>	<b>Combined<sup>a</sup></b>
Overall, how would you rate your skills with technology?	7.14 (1.07) 5-8	7.14 (2.19), 3-10	6.56 (1.76), 4-10	6.91 (1.76), 3-10
What would you give out of 10 for your child's experience of the application?	7.29 (1.80), 5-10	7.86 (1.57), 6-10	6.44 (2.60), 2-10	7.13 (2.10), 2-10
What would you give out of 10 for your experience of the application?	8.00 (2.16), 5-10	8.57 (1.27), 7-10	7.89 (1.83), 5-10	8.13 (1.74), 5-10
What would you give out of 10 for the benefits to your child in using the application?	7.43 (1.98), 5-10	9.00 (1.27) 7-10	7.11 (2.89), 2-10	7.73 (2.31), 2-10
What would you give out of 10 for benefits to you in using the application?	7.29 (2.75), 2-10	9.00 (1.41), 7-10	8.11 (1.83), 4-10	8.13 (2.07), 2-10
What would you give out of 10 for the application's usability (how easy or hard was it to use the application) with 10 being very easy and 0 being very difficult?	8.57 (1.99), 5-10	9.29 (1.25), 7-10	7.89 (1.69), 5-10	8.52 (1.70), 5-10

<sup>a</sup> Mean; (SD); range

## **5.5 Discussion**

In this study, parents living in regional areas who completed three months of the intervention as part of an RCT examining the effectiveness of the TOBY app, an ICT-based intervention, were interviewed to evaluate the intervention's appropriateness (Parsons et al., 2019). To the authors' knowledge, this is the first study to apply an appropriateness framework to an ICT-based intervention for families of children with ASD. Further, this is the first study to investigate the experience of using an ICT-based intervention for families of children with ASD living in regional areas. The thematic analysis generated the core theme — the TOBY app was not a panacea for all their children's barriers. Importantly, this core theme reflects the heterogeneity in parents' perceptions in the outcomes for their child with ASD who used the intervention, not the TOBY app's failure to address all barriers in all children, which is not the TOBY app's intended function. Furthermore, collective responses from parents indicated that the TOBY app was more appropriate for some children with ASD, but less so for others. Parents suggested that the TOBY app should be just one part of the therapy landscape, and cannot and should not replace face-to-face therapy, but complement it. This finding reinforces similar findings of the seminal work by Silver and Oakes (2001).

### **5.5.1 Applying the core themes to an appropriateness framework**

#### **5.5.1.1 Intervention as a positive experience, relevance and importance**

Parents reported mixed results in their experience with the TOBY app. Most parents stated the TOBY app was straightforward to use, with clear instructions and easy navigation. These findings are consistent with previous studies investigating the user experience for the TOBY app (Rogerson et al., 2018; Whitehouse et al., 2017). Despite this, some parents reported some issues with the TOBY app that tainted their experience: 1) it was challenging to get their children to engage with the app for 20 minutes per day; 2) a limited

ability to choose and control the activities completed on the app; 3) the manifestation of problem behaviours in their children associated with using the TOBY app; and 4) the need for ongoing support from therapists, which they did not receive as part of this research project. Parents reported negative experiences with the TOBY app may have limited the dosage and fidelity of the intervention, thus reducing the benefits of the ap. Collectively, these findings partially support the TOBY app is appropriate from a user perspective; however, more need to be done to improve this experience for both parents and children.

These findings highlight the need for developers and researchers of ICT-based interventions for children with ASD to pay particular attention to the user experience of both the parents and the children. That is, ICT-based interventions for children with ASD should be engaging, easy-to-use, responsive to children's developmental level, and include customisation options for parents to better individualise the intervention to their children's needs and intrinsic motivations (Whyte, Smyth, & Scherf, 2015). The heterogeneity between children with ASD creates the need for developers to give users greater choice and control over their experiences, such as customisable pictures and activities, to improve outcomes through increased engagement, dosage and treatment fidelity. This finding builds on the work by Allen, Hartley, and Cain (2016) who recommend ICT-based interventions should have the ability to create and integrate customised visual inputs to improve the child's language and social skills.

Current literature supports the inclusion of gamification elements in ICT-based interventions through storylines and goal-directed learning to enhance motivation and contextualise learning (Baranowski, Buday, Thompson, & Baranowski, 2008; Whyte et al., 2015). Further, by parents having increased control over their children's experience through increased customisation, the children using the intervention should have a more

contextualised learning experience. An increased contextual learning experience could not only increase engagement in the child, but may also increase the likelihood of transference of learnt skills into real-life contexts; a well-known challenge in ASD interventions (Kourkoulou, Leekam, & Findlay, 2012; Ramdoss et al., 2012).

Parents in the study acknowledged the relevance of the TOBY app, with all participants expressing their desire to help their children overcome their developmental challenges. Additionally, all parents interviewed would recommend the TOBY app to a friend, even if they felt it was not beneficial for their children, indicating they believe its utility and relevance for helping children with ASD. Therefore, the participants support the appropriateness of the TOBY app in relation to its relevance and importance to both them and their children.

#### **5.5.1.2 Intervention is beneficial**

Parents reported varying levels of benefit for their children across a broad range of skill areas, indicating the TOBY app was not superior at developing any one skill over another, but did have utility in increasing the amount of EIBI their child received. However, the sub-theme '*The TOBY app is not for everyone*' suggests the TOBY app is not appropriate for every child with ASD. Parents reported the TOBY app allowed them to become better at helping their children by increasing their knowledge and skills with ASD, including ASD interventions, and by understanding their children's needs more. Specifically, parents reported an increased understanding of their children's strengths and weaknesses, while at the same time improving the EIBI skills the app was teaching them. This is a key finding in this study and is congruent with the outcomes in remotely-delivered or parent-mediated intervention effectiveness studies for families of children with ASD (Heitzman-Powell, Buzhardt, Rusinko, & Miller, 2014; Hutton & Caron, 2005; Vismara, McCormick, Young,

Nadhan, & Monlux, 2013; Wacker et al., 2013). With increased knowledge, skills, self-efficacy and understanding of their children's needs, parents are better poised to become more skilled as active agents of change in their children's development (McConachie & Diggle, 2007; Solish & Perry, 2008). Findings from this study suggest the TOBY app is appropriate for parents, key agents in the delivery of the TOBY app, but should be framed with consideration of the previous finding — that the TOBY app is too rigid and lacks the individualisation that face-to-face therapy can provide.

Lastly, it should be noted that the perceived benefits from parents could result from parental expectations and placebo effects (Fageera et al., 2018; Masi, Lampit, Glozier, Hickie, & Guastella, 2015). Moreover, given the lack of significant changes in visual motor, imitation, receptive language and social skills in the effectiveness trial, interpretation of this finding should be considered with due caution. Overall, these findings partially support the TOBY app's appropriateness as a beneficial intervention; however, it was evident that the TOBY app did not benefit all participants.

#### **5.5.1.3 Intervention as social and ecologically valid intervention and change and continuation**

The core theme '*The TOBY app is just one piece of the jigsaw*' supports the partial social and ecological appropriateness of the TOBY app. For some parents, it was achievable to complete the desired 20 minutes of therapy once-per-day; for other parents this was not an achievable goal. Busy family lives, school commitments, other therapy appointments, and a lack of time were often cited as barriers. These findings are similar to those reported in other studies regarding the barriers to completing parent-delivered or Internet-based interventions (Mackintosh, Goin-Kochel, & Myers, 2012; Sinclair, Holloway, Riley, & Auret, 2013). Parents in this study reported that ongoing support from a therapist would be beneficial when using the TOBY app, indicating ongoing support would improve the



overall appropriateness of the intervention. However, the increased support may have the unintended consequence of reducing the feasibility of the intervention. This finding builds on previous studies reporting therapist support, initial training for parents to use the intervention, and knowledge sharing increase parents' satisfaction and sense of competence in the delivery of interventions (Allan et al., 2018; Allen et al., 2016; Foster, Dunn, & Lawson, 2013).

Some parents stated that the TOBY app was useful for them living in regional areas as it could help reduce, but not replace, the number of face-to-face therapy sessions required. Thus, decreasing the distance travelled to access these services and increasing the ecological appropriateness of the intervention. Service delivery models incorporating telehealth and ICT-based interventions are emerging as viable and feasible intervention delivery methods for families of children with ASD experiencing service access issues due to geographical distance (Antezana, Scarpa, Valdespino, Albright, & Richey, 2017). The findings from this study support the potential of ICT-based interventions, including the TOBY app, for clinicians and families living regional areas in alleviating some of the considerable economic burden associated with accessing the appropriate services (Hoogsteen & Woodgate, 2013; Horlin, Falkmer, Parsons, Albrecht, & Falkmer, 2014).

A number of parents reported continued use of the skills and strategies learnt throughout the intervention periods, while others had ceased entirely. Notably, some parents reported improvements of their children in daily living skills that they directly attribute to using the TOBY app, such as bath and mealtime routines, achieved throughout the intervention period had been maintained up to 12 months after cessation of the TOBY app use with the ongoing use of strategies learnt. Therefore, there is partial support for the appropriateness of the TOBY app as an intervention that promotes change and the continuation of learnt

skills in children with ASD and their parents.

### **5.5.2 Limitations**

This study has some notable limitations. Participants in this study were recruited solely from participants in the RCT by Parsons et al. (2019). Recruiting only from this source could skew respondents to those who already perceive the relevance of the TOBY app, therefore contributing to selection bias. The level of support provided to the participants in the RCT was restricted due to available resources of the researchers and to increase the ecological validity, based on the assumption that, in real life, families can download the app and use it without any support.

In this study, all participants were mothers with only one living in a low socioeconomic area based on the SEIFA index (Pink, 2011). Hence, generalisation of study findings to fathers, lower socioeconomic populations, and parents living in major cities may be limited. However, participants in this study are likely to be representative of those who are most likely to use the TOBY app and other ICT interventions, given mothers in families of a child with ASD are more likely to adopt primary caregiver roles, such as delivering home-based interventions like the TOBY app, compared to families without children with ASD (McAuliffe, Cordier, Vaz, Thomas, & Falkmer, 2017; Nealy, O'Hare, Powers, & Swick, 2012)

Finally, although the primary researcher – the interviewer – declared no conflict of interest with the TOBY app at the beginning of each interview, due to resourcing issues, the primary researcher conducted 20% of the assessments in the RCT. Participants in this study could perceive a conflict of interest and tailor their responses accordingly.

### **5.5.3 Implications and future research**

This study illustrates that ongoing support is a necessary component in the provision of

ICT-based interventions for families living in regional and remote areas and should be a consideration for practitioners working with families from these areas. Future research into the role of ICT-based interventions as a complement to telehealth interventions to improve accessibility and reduce the economic impact for families who need to travel vast distances to gain access to services is warranted.

Parents expressed the need for ongoing support when using the TOBY app. Researching the experiences of the TOBY app from therapists' perspectives could provide valuable insight. Convergence of therapists' and parents' data may provide further strategies for the ongoing development and implementation to improve therapy outcomes for children with ASD using the TOBY app and other ICT-based interventions. Further investigation into the optimal level of support provided to parents implementing the TOBY app should be considered. Given the increasing demand for therapy resourcing, having increased knowledge regarding the level of support required to ensure parents can effectively deliver the intervention will improve the feasibility of ICT-based interventions. Knowing the optimal level of support to provide could also help clinicians improve the experience of the parents and benefit of the children from using the TOBY app.

Furthermore, the subtheme '*The TOBY app is not for everyone*' suggesting the TOBY app is not appropriate for every child is congruent with other studies investigating predictors for symptoms change in children with ASD. That is, due to the highly heterogeneous nature of ASD, children with ASD respond very differently to the same interventions, and more research is required to ascertain "what works for whom and why" (Hudry et al., 2018; Vivanti, Prior, Williams, & Dissanayake, 2014). Lastly, this study used maximum variation purposive sampling that included low, medium and high users of the TOBY app to obtain a rich and overarching narrative based on information related to both the enablers and the

barriers in using the intervention. This was done by gaining insights from participants with varying levels recorded use of the app to evaluate the appropriateness of the TOBY app. Between-group comparisons were not conducted to identify factors influencing dosage and adherence as the inductive nature of thematic analysis does not allow for making meaningful inferences to the broader population outside of this sample. Future research could investigate the factors (including predictor variables) that influence dosage and adherence to the TOBY app intervention using both qualitative and quantitative research methods.

## **5.6 Conclusion**

Findings from this study partially support the appropriateness of the TOBY app for children with ASD and their parents who live in regional Australia. Thematic analysis of interviews of parents who used the TOBY app as part of an effectiveness study identified the core theme that the TOBY app is not a panacea for the challenges associated with ASD. Collectively, parents reported that that the TOBY app was appropriate for some children and not others, and should be used to complement other therapies and not in isolation. Ongoing support from therapists, increased customisation through more choice and control, and a focus on user experience was highlighted by parents as strategies that may improve the overall appropriateness of the TOBY app.

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## Chapter 6      Maintenance of intervention effects for the Therapy Outcomes By You application

Chapter 6 describes a follow-up study for children with autism spectrum disorder living in regional Australia who used the Therapy Outcomes By You application for three months in the randomised controlled trial to determine if children with autism spectrum disorder maintained or continued to improve their language and social communication skills after at least 12 months post-intervention. This chapter is currently under review in the Journal of Autism and Developmental Disorders.

The spelling and wording contained within this chapter are that of the submitted manuscript.





# Journal Manuscript 5

A twelve-month follow-up of an information communication technology delivered intervention for children with autism spectrum disorder living in regional Australia.

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## **6.1 Abstract**

This study investigated the long-term follow-up of an Information Communication Technology based intervention, the Therapeutic Outcomes By You (TOBY) application, for children with Autism Spectrum Disorder (ASD) living in regional Australia. Fifteen participants who completed a three-month randomised controlled trial of the TOBY were assessed at least 12 months post-intervention to determine the maintenance or continued improvement of their language and social communication skills. Findings demonstrate the receptive language, social skills, pragmatic language and playfulness of children with ASD improved during the three-month intervention period and were maintained at least 12 months after ceasing the TOBY app intervention.

**Keywords:** Information technology, early intervention, parent training



## 6.2 Background

Autism spectrum disorder (ASD) is a pervasive life-long neurodevelopmental condition characterised by persistent repetitive patterns of behaviour, language communication difficulties and social interaction impairments (American Psychiatric Association, 2013). Early Intense Behavioural Interventions (EIBI) have been reported as effective in reducing the core features of ASD and have been mooted as long-term cost-effective interventions due to the improvements in skills resulting in a reduced need for programs and supports as the child matures (Dawson et al., 2010; Matson & Konst, 2013; Oono, Honey, & McConachie, 2013; Ramdoss et al., 2012). However, the overall cost-effectiveness of an EIBI relies on the assumption that the child maintains the skills learnt during the intervention period and beyond. Moreover, the long-term maintenance of skill acquisition in EIBI intervention studies is seldom investigated (Matson & Konst, 2013).

Current best practice guidelines for EIBI require the child to receive at least 25 hours of EIBI per week to improve skills in imitation, joint attention, play skills, and both receptive and expressive language (Prior & Roberts, 2012). Despite its long-term benefit, this level of intensity often places significant short-term financial and psychological strain on the family,; a consideration that is even more pronounced when families need to travel long distances to access services (Bailey, Hebbeler, Scarborough, Spiker, & Mallik, 2004; Horlin, Falkmer, Parsons, Albrecht, & Falkmer, 2014; McAuliffe, Vaz, Falkmer, & Cordier, 2016; Prior & Roberts, 2012; Sim et al., 2018). With the high financial, psycho-emotional and access challenges experienced by families of children with ASD, there is a need to assess the long-term impact of EIBI to ensure the intervention has long-term benefit for the child (Matson & Konst, 2013).

The United Kingdom Medical Research Council (UKMRC) guidance for the development

of complex intervention recommends long-term follow-up of participants to determine if short-term changes persist and while these studies are uncommon, they are highly informative (Campbell et al., 2000; Craig et al., 2008). Further, if interventions do not demonstrate maintenance, then time and resources invested into teaching skills or implementing interventions with poor long-term efficacy could potentially be squandered (Alper & Raharinirina, 2006).

Preliminary evidence supporting the efficacy of ICT-based interventions for children with ASD is continuing to develop with robust randomised controlled trials and systematic reviews supporting their use to improve social, emotional and communication skills (Parsons, Cordier, Lee, Falkmer, & Vaz, 2019; Ramdoss et al., 2011; Ramdoss et al., 2012; Wainer & Ingersoll, 2011; Whitehouse et al., 2017). However, few studies have included long-term follow-up in their design to assess the maintenance of skills. The Therapeutic Outcomes By You application (TOBY app) is an early intervention iPad application and intervention tool based on Applied Behavioural Analysis (ABA) principles and EIBI guidelines, which was specially developed by a multidisciplinary team comprising of psychologists, speech pathologists and computer scientists for children aged two to six with ASD (Moore et al., 2015; Parsons et al., 2019; Venkatesh, Phung, Duong, Greenhill, & Adams, 2013).

The TOBY app targets the following skills areas: 1) receptive and expressive language; 2) sensory awareness; and 3) imitation and social interaction skills such as joint attention and gestures (Moore et al., 2015; Parsons et al., 2019). The TOBY app was designed to supplement existing therapy, not replace it. The application is a low-cost intervention and can be easily accessed through the Apple App store (AUD\$25.99) (Moore et al., 2015). Although not designed for this purpose, parents can use the TOBY app at home

independent of any clinician input. The developers of the TOBY app claim its uniqueness is in how the intervention teaches a parent how to teach the child (Venkatesh et al., 2013).

One pilot study and two RCTs have evaluated the effectiveness of the TOBY app to date.

The pilot study by Moore et al. (2015) reported that the TOBY app delivers reliable and accurate feedback, with the difficulty level being appropriately matched to the child's abilities. In their RCT, Whitehouse et al. (2017) demonstrated the effectiveness of the TOBY app in improving fine motor skills and visual motor skills; however, concluded the TOBY app does not reduce autism symptom severity levels in children aged two to six.

The second RCT by Parsons et al. (2019), from which the participants from this study were recruited, investigated the effectiveness of the TOBY app for children with ASD with a developmental age of two to six years who live in regional areas. The authors reported statistically significant improvements in expressive language in children with ASD between the intervention group, who were instructed to use the TOBY app for 20 minutes per day over a three-month period in addition to therapy-as-usual, and the control group who received therapy-as-usual without TOBY app use. Furthermore, improvements in receptive language, pragmatic language and social communication skills were detected within the intervention group participants ( $n = 59$ ) when measured pre-post over three months, suggesting skill acquisition (Parsons et al., 2019). Although findings from the RCT effectiveness studies provide limited evidence the TOBY app is an effective intervention to improve the receptive language, social communication, fine motor and visual motor skill in children with ASD immediately post-intervention, a long-term follow-up study with participants to investigate the maintenance of these skills following use of the TOBY app has not yet been conducted.

To date, there is no evidence of maintenance effects for the TOBY app. Previous studies

have reported the maintenance of skills in children with ASD following psychosocial interventions similar to those taught in the TOBY app, such as social and language skills (Jones, Carr, & Feeley, 2006; Kasari, Gulsrud, Wong, Kwon, & Locke, 2010; Wert & Neisworth, 2003). In a systematic review investigating the quality of behaviourally-based intervention to improve social interaction skills for a child with ASD, Camargo et al. (2014) reported that of 15 studies investigating the maintenance of social interaction skills in children with ASD, 13 studies showed children retained at least one of the intervention's target skills. Additionally, improved maintenance of learnt skills is achieved if the intervention is implemented in the context where the child will use the skill (Camargo et al., 2014). However, there are no studies to these author's knowledge that have investigated the maintenance of psychosocial skills following an ICT-based intervention in children living in regional areas.

This paper aims to report on a follow-up study of children with ASD living in regional Australia who used the TOBY app for three months in the RCT by Parsons et al. (2019) to determine if participants maintained or continued to improve their language and social communication skills after at least 12 months post-intervention. The study also aimed to explain the maintenance of skills and ongoing use of the TOBY app from the perspectives of the parents who participated in the RCT.

## **6.3 Methods**

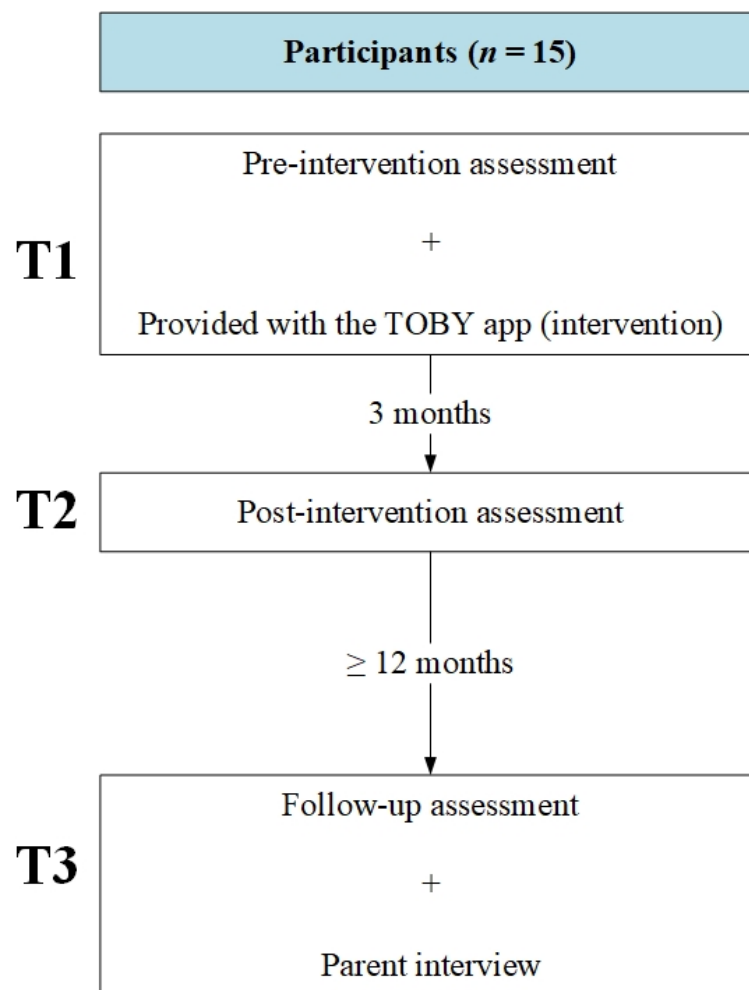
### **6.3.1 Study design and recruitment**

This study used a single-site cohort design, with data collected at baseline (T1), post-intervention (T2) and follow-up at 12 months post-intervention (T3) (see Figure 6.1).

Participants were sampled from a larger sample who participated in an effectiveness study using an RCT design and had used the TOBY app for a minimum of three months (Parsons



et al., 2019). We used purposive sampling to recruit participants from the RCT trial into this study to ensure maximum variation. Participants were invited based on recorded app use from the lowest, middle and highest segments from the RCT study across the duration of the study. The rationale was to gather data from participants with varying levels of engagement in the RCT with the aim to obtain a representative sample to increase the generalisation of findings. Back-end server data automatically generated during app use were accessed and analysed as an objective measurement of usage. Usage was measured as a summation of participant ranking based on three components: 1) time on the app; 2) number of items attempted; and 3) number of items completed. Fifteen families with a child with ASD from the RCT agreed to participate in this study.



**Figure 6.1 Outline of study procedure**

### **6.3.2 Participants**

#### **6.3.2.1 Inclusion and exclusion criteria**

All children in the study had a diagnosis of ASD as determined by a team of qualified health professionals using the *Diagnosis and Statistical Manual of Mental Disorders (DSM-5)* (American Psychiatric Association, 2013). The children had a developmental age between two and six years and resided in areas outside of major cities in Western Australia as defined by the Australian Standard Geographical Classification System (Australian Bureau of Statistics, 2011). Participants were recruited through key ASD service providers including paediatricians, general practitioners (GPs), allied health clinicians (e.g., speech pathologist, occupational therapists, psychologists), parent support groups, community forums, media advertisements and snowballing techniques from the south-west region of Western Australia. All participants were invited to conduct the interview via the telephone. Children with existing comorbidities commonly occurring with ASD (e.g., anxiety, attention deficit hyperactivity disorder) were included provided ASD was their primary diagnosis. If the parent consented, a face-to-face assessment was arranged at the convenience of the parents for the long-term follow-up. Informed written consent was obtained from all participants at the follow-up assessment. All assessments for the long-term follow-up were completed in the participants' home.

Children were excluded from the study if they had a physical disability that prevented them from engaging in the recommended 20 minutes of therapy per day with the TOBY app. Further, children were excluded from the study if they had non-idiopathic cases of ASD including genetic disorders, such as Rett's syndrome and Fragile X syndrome (Luyster, Kadlec, Carter, & Tager-Flusberg, 2008). Informed consent was obtained prior to participation in the study. Ethical approval was obtained from the Curtin University Human Research Ethics Committee (HR123/2014).

### **6.3.2.2 Demographic information**

Demographic information was collected from all participants for: 1) child age; 2) child gender; 3) maternal education; 4) paternal education; and 5) regionality. Socioeconomic status (SES) was determined using the Socio-Economic Index for Areas (SEIFA) deciles. The SEIFA deciles are developed by the Australian Bureau of Statistics to rank areas according to their relative socio-economic advantage and disadvantage based on information from a five-yearly census (Australian Bureau of Statistics, 2011). The SEIFA deciles are represented using a ten-point numerical scale, with a higher SEIFA score representing a higher SES status. Pink (2011) reports that a score of 1 to 3 reflects low SES, with 4 to 10 representing medium to high SES.

To define regionality, the Australian Geographical Classification System (AGCS) was used. The AGCS divides Australia into broad regions for comparative statistical purposes (Australian Bureau of Statistics, 2011). The ASGC remoteness structure is classified into five remoteness areas (RAs) (Australian Bureau of Statistics, 2011). The categories include: 1) major cities; 2) inner regional; 3) outer regional; 4) remote; and 5) very remote. RAs are based on road distances to the nearest service centres and average scores are calculated using the Accessibility/Remoteness Index of Australia (ARIA +) grid, which is a one square kilometre grid covering all of Australia (Australian Bureau of Statistics, 2011; McAuliffe et al., 2016).

### **6.3.3 Instruments**

A series of standardised repeated measures were administered at baseline (T1), post-test (T2) and at follow-up (T3) to assess the effectiveness of the TOBY app. These measures included: 1) Mullen Scales of Early Learning (MSEL) (Mullen, 1995); 2) Symbolic Play Test (SPT) (Lowe & Costello, 1988); and 3) Communication and Symbolic Behavior

Scales (CSBS) (Wetherby & Prizant, 2002). Additionally, a 20-minute video of spontaneous interaction with a neuro-typical person was obtained at all data collection time points. Two standardised observation measures were used to analyse the video data; 1) Pragmatic Observation Measure (POM); and 2) Test of Playfulness (ToP) (Bundy, 2004; Cordier et al., 2019). Two experienced occupational therapy clinicians trained and calibrated in the POM and ToP analysed the video footage and were blinded to all aspects of the study.

The MSEL (visual reception, and expressive and receptive language skills) and CSBS (imitation and social skills) were used as the primary outcome measures in the intervention. The POM, SPT and ToP were administered as secondary outcomes measures, as hypothesised skill acquisition resulting from the TOBY app use could be developmental precursors to pragmatic language, symbolic play skills, and play. Further, these assessments were all administered in the RCT where the participants were recruited; therefore, to accurately measure maintenance, the use of the same instruments was warranted.

#### **6.3.3.1 Mullen Scales of Early Learning (MSEL)**

The MSEL comprises five sub-scales that assess a child's abilities in visual reception, gross motor functions, fine motor skills, receptive language and expressive language (Mullen, 1995). The measure is a comprehensive measure of cognitive functioning for infants and pre-school children, from birth to 68 months. For this study, the gross motor scale was not administered as it was not age-appropriate. The MSEL is deemed to be valid and appropriate in assessing children with ASD (Akshoomoff, 2006). Developmental quotients (developmental age divided by chronological age, multiplied by 100) were derived for the four sub-scales given some participants in the study achieved T-Scores at or

below 20, thus representing 3 or more standard deviations below the mean. Reliability estimates are moderate (Cronbach's alpha values range 0.75 - 0.83) with a composite median value of 0.91. Inter-rater reliability was very high, with a range of 0.91 to 0.99 (Mullen, 1995).

#### **6.3.3.2 Communication and Symbolic Behavior Scales Developmental Profile Caregiver Questionnaire (CSBS)**

The CSBS is a caregiver-reported standardised assessment designed to measure: 1) speech skills, such as sounds and words'; 2) social-affective skills, such as emotion and eye gaze, communication, and gestures; and 3) symbolic abilities, such as understanding and object use of children (Levy et al., 2003). The caregiver questionnaire was used in this study.

The measure has moderate to strong concurrent validity ( $r = 0.59 - 0.61$  and  $0.65 - .071$ ) with reference to the one-page caregiver report checklist and behaviour sample (face-to-face) of the same assessment, indicating its validity as a tool for measuring the communication and language skills of young children (Wetherby, Allen, Cleary, Kublin, & Goldstein, 2002). The CSBS has moderate to strong test re-test reliability ( $r = 0.64 - 0.91$ ) for the checklist questionnaire and behaviour sample. The scales also exhibit high internal consistency (Cronbach's alpha range 0.86 - 0.92) (Wetherby & Prizant, 2002).

#### **6.3.3.3 Pragmatic Observation Measure (POM)**

The POM is a 27-item observer rated instrument of pragmatic aspects of language based on direct observation (Cordier et al., 2019). The POM has evidence for excellent inter-rater reliability (0.89) and internal consistency (Cronbach's alpha  $> 0.98$ ). The measure has excellent criterion validity (0.95), good construct validity (0.55 - 0.77) and high responsiveness to change (sensitivity = 79.7%; specificity = 89.6%).

#### **6.3.3.4 Test of Playfulness (ToP)**

The ToP measures combined presence of four the elements of play: internal control, freedom from unnecessary constraints of reality, intrinsic motivation, and framing (the ability to give and read social cues) to measure the concept of playfulness. The measure is a 29-item observer-rated instrument suitable for children between 6 months and 18 years. Based on raw scores, the ToP has moderate test-retest reliability with significant intra-class correlation 0.67 ( $p < 0.01$ ) (Bundy, 2004). Further, the ToP has evidence for excellent inter-rater reliability, supported by data from 96% of raters who fitted the expectations of the Rasch model. The measure demonstrates good construct validity with data from 93% items and 98% of people fitting Rasch expectations (Bundy, 2004).

#### **6.3.4 Open-ended questions**

After the assessment, participants were asked a series of open-ended questions lasting between 5 – 15 minutes to provide further explanation regarding the continued use and maintenance of skills learnt while using the TOBY app. The qualitative data was explanatory in nature with the intention of clarifying quantitative responses and not conduct in-depth interviews. All interviews were recorded on a digital voice recorder and were transcribed verbatim.

#### **6.3.5 Intervention**

The TOBY app comprises the following three types of tasks: solo, partner, and natural environment tasks (NET). Children begin the intervention with activities at their current level of functioning and progress through the curriculum at their own rate of development and ability. Solo tasks are completed by the child independently, while partner tasks are undertaken with caregivers' assistance for recording responses or providing prompts and stimuli (Venkatesh et al., 2013). The NET tasks are performed separately from the iPad

with caregiver support and are integrated into daily life to encourage generalisation of skills learnt during solo and partner tasks. Responses to each task are inputted into TOBY app, and a syllabus of future tasks is tailored for the child. This intervention can be delivered in the home by the parent or caregiver, without the direct involvement of health professionals (Moore et al., 2015; Venkatesh et al., 2013). For more information about the TOBY app, refer to the intervention description in the published RCT of the intervention (Parsons et al., 2019).

#### **6.3.6 Data analysis**

Outcome measure data were managed and analysed using SPSS<sup>®</sup> 25 (IBM Corp., 2016). Descriptive statistics were used to analyse the demographic data of the sample. Linear mixed models were used to measure change over time at baseline (T1), post-intervention (T2) and follow-up (T3) using the fixed effect of time with an autoregressive covariance matrix to define the within-subject error, using coefficients estimated via maximum likelihood. Post-hoc pairwise comparisons were conducted from T2 to T3 to determine maintenance of the intervention. A linear mixed model was used given its suitability for modelling the influence of nonlinear individual differences across time and an approach recommended for the evaluation of psychological clinical trials (Hamer & Simpson, 2009; Krueger & Tian, 2004).

Analysis of open-ended responses was guided using the thematic analysis approach outlined by Braun and Clarke (2006). The approach is more descriptive when analysing the data, relying less on interpretation by the researchers and more on the description of experiences by participants (Creswell, 1998). Thematic analysis allows the researcher to highlight the similarities and differences across the data set, provide a large amount of flexibility and is useful when using large bodies of data (Braun & Clarke, 2006). NVivo<sup>®</sup>

(version 12) was used as the data management software. A clear audit trail using thematic analysis was maintained throughout the process. Line-by-line coding and categorisation were completed by the first author using the transcriptions of the interviews. The data was then analysed for trends and patterns of word use, frequency, their relationship and structures of discourse of communication. All interviews were completed by the same researcher, the fourth author, to enhance consistency. Summaries of the interviews were sent to the participants for member checking to ensure the accuracy of their responses prior to thematic analysis. All inconsistencies were corrected prior to commencing thematic analysis. Finally, throughout the data analysis, process interpretations were cross-checked over several research meetings between the first and fourth authors until consensus was reached.

## **6.4 Results**

### **6.4.1 Participants**

Nineteen participants were invited to participate in the follow-up assessment. Four parents declined to participate, thus, 15 parents and their children were included in the study. All parents who answered the open-ended questions in the follow-up study were mothers. Refer to Table 6.1 for the demographic information of the participants.

### **6.4.2 Areas showing continued improvements during observation period of three months**

Mean follow-up time post intervention was 14.5 months (range = 12 to 18, SD = 1.85). Pre-post intervention analysis using the three time points of the participants demonstrated statistically significant improvements in the: 1) MSEL receptive language subscale; 2) social, symbolic and speech subscales and the total composite score of the CSBS; 3) POM; and, 4) ToP (see Table 6.2). Post hoc analysis using pairwise comparisons demonstrated statistically significant improvements between T1 and T2 for MSEL receptive language



subscale ( $p = .014$ ), CSBS social communication subscale ( $p = .003$ ), CSBS symbolic subscale ( $p = .001$ ), CSBS speech subscale ( $p < .001$ ), CSBS total composite score ( $p < .001$ ), POM ( $p = .002$ ), and ToP ( $p = .007$ ). Importantly, no significant differences were detected between T2 and T3 time points for the outcomes MSEL receptive language subscale ( $p = .054$ ), CSBS social communication subscale ( $p = .160$ ), POM ( $p = .809$ ), and ToP ( $p = .172$ ); indicating that the participants' skills did not improve significantly after ceasing the intervention, however, they also did not regress in these skill areas, thus demonstrating maintenance.

#### **6.4.3 Comparison between randomised controlled trial and follow-up study results**

While improvements in expressive language were detected between the intervention and control groups in the RCT, the comparison of the intervention and control groups in this follow-up study could not be completed due to the waitlist design of the RCT. That is, all participants regardless of allocation to the intervention or control group for the RCT received the TOBY app intervention. Notably, the final time point in the study by Parsons et al. (2019) was completed at three months (T3) compared to the longer follow-up time point at 12 months (T4) in this study. See Table 6.3 for a comparison of results from the RCT study with the results from this study.

**Table 6.1 Child and family characteristics at the 12-month follow-up time point**

Participant Characteristics		Follow-up ( <i>n</i> =15)
Child Age in months	Mean (SD)	79.07 (22.33)
Child Gender	Male	11 (73.3%)
	Female	4 (26.3%)
Number of Children with ASD	1	12 (80%)
	2 or more	3 (20%)
Maternal Education	Diploma or Below	11 (73.3%)
	Bachelor Degree or higher	4 (26.7%)
Paternal Education	Diploma or Below	12 (80%)
	Bachelor Degree or higher	3 (20%)
Population density of area*	Inner Regional	13 (86.7)
	Outer Regional	2 (13.3%)
	Remote	0 (0%)
	Very Remote	0 (0%)
SEIFA Decile	Mean (SD)	6.53 (1.68)

*Note.* \*Based on the Australian standard geographical classification system (Australian Bureau of Statistics, 2011). The categories include major cities, inner regional, outer regional, remote, and very remote based on a number of variables including population size and distance by road to service centres.

SEIFA: Socio-economic index for areas

**Table 6.2 Linear mixed model - means and standard error for all baseline, outcome and follow-up measures pooled**

		Baseline (T1)	3-Month (T2)	Follow-up (T3)	F- score	p value
		(n=15)	(n=15)	(n=15)		
MSEL	Visual Reception	72.6 (6.01)	79.8 (6.1)	70.9 (6.01)	2.347	.118
	Fine Motor	66.0 (5.24)	71.1 (5.24)	65.6 (5.24)	2.446	.111
	Receptive Language	62.5 (6.34)	72.7 (6.45)	70.3 (6.93)	3.725	.042*
	Expressive Language	62.7 (6.39)	66.1 (6.55)	67.3 (6.39)	.771	.476
SPT	Age Equivalent <sup>§</sup>	31.9 (1.40)	31.4 (1.39)	31.1 (1.38)	.279	.759
CSBS	Social Domain	31.1 (1.76)	34.8 (1.72)	36.4 (1.74)	10.730	.001*
	Speech Domain <sup>§</sup>	31.7 (1.36)	36.8 (1.29)	39.3 (1.32)	14.072	< .001*
	Symbolic Domain	38.5 (1.30)	42.9 (1.22)	46.5 (1.25)	13.659	< .001*
	Total Composite	101.23 (3.50)	114.6 (3.33)	122.1 (3.34)	26.678	< .001*
POM		11.6 (8.60)	38.8 (8.60)	36.8 (8.60)	5.774	.008*
TOP		47.24 (3.97)	59.49 (3.97)	53.6 (3.97)	4.316	.026*

\*Significant difference ( $p$ -value > .05)

<sup>§</sup>Ceiling Effect

MSEL: Mullen Scales of Early Learning

SPT: Symbolic Play Test

CSBS: Communication and Symbolic Behaviour Scale

POM: Pragmatic Observation Measure

TOP: Test of Playfulness

**Table 6.3 Comparison of maintenance at three months (T3) and at greater than 12 months (T4)**

		RCT study (n = 59)			Follow-up study (n = 15)	
		Between-group comparison (T1 – T2) <sup>a</sup>	Pre-post intervention (T1- T2) <sup>b</sup>	Maintained skills (T2 – T3)	Pre-post intervention (T1 – T2)	Maintained skills (T2 – T4)
MSEL	Visual Reception§					
	Fine Motor					
	Receptive Language		•	•	•	•
	Expressive Language	•				•
SPT	Age Equivalent§					
CSBS	Social Domain		•	•	•	•
	Speech Domain <sup>§</sup>				•	†
	Symbolic Domain		•	•	•	†
	Total Composite <sup>§</sup>	×	×		•	†
POM			•	•	•	•
TOP					•	•

<sup>a</sup> Intervention vs Control

<sup>b</sup> Pooled participant data

<sup>§</sup> Ceiling Effect

† Improved significantly from T2 to T4

× Not reported in RCT study

MSEL: Mullen Scales of Early Learning

SPT: Symbolic Play Test

CSBS: Communication and Symbolic Behaviour Scale

POM: Pragmatic Observation Measure

TOP: Test of Playfulness

#### **6.4.4 Parent responses**

In addition to using standardised outcome instruments to measure the developmental outcomes of the children who participated in study 12 months after the intervention, the study sought to seek parents' opinions on the continued use of the app and the maintenance of skills learnt while using the TOBY app. Thirteen out of 15 parents reported no longer using the app or using the app 'very little' after the initial three-month intervention period. Parents reported a number of reasons as to why they had stopped using the application since the trial ceased, with the main findings summarised in Table 6.4. A lack of time and a loss of interest from their child in the application were cited as the main reasons. Other key findings from the interviews were the TOBY app's level of difficulty became too easy for their child, which contributed to a loss of interest for both the parents and children and discontinued use was due to parents changing the therapy goals for their child towards skills that the TOBY app did not target. Finally, 12 out of 15 parents reported their child had maintained at least one skill they developed while using the TOBY app, including receptive language, social communication or daily living skills — thus supporting the quantitative findings of maintenance in these skill areas. Triangulation by analysing the individual changes for each child from T2 to T3 for each outcome measure confirmed these parent reports.

**Table 6.4 Thematic representations of parent interviews on continued use of the TOBY app and their child's maintenance of life skills**

Topic	Themes	Common responses
Discontinued use of the TOBY app	Lack of time	<ul style="list-style-type: none"> <li>• Difficult to fit in the 20 minutes a day</li> <li>• Caring responsibilities to other children</li> <li>• Child beginning school</li> <li>• Lifestyle factors of returning to work or having another child.</li> <li>• Extra-curricular activities</li> <li>• Tedious to read the instructions and takes too much time.</li> </ul>
	Skills taught by the TOBY app were no longer the goal	<ul style="list-style-type: none"> <li>• Wanted to work on social behaviour which mother thinks are not covered by TOBY</li> <li>• Needed to focus on other forms of therapy to build social skills and sensory related issues.</li> <li>• Needed that time and effort to focus on other therapies.</li> </ul>
	Child lost interest	<ul style="list-style-type: none"> <li>• Could not focus on the TOBY app for 20 minutes a day.</li> <li>• As the tasks were too easy, the child got bored.</li> <li>• Not enjoying application</li> <li>• The child got frustrated at the voice on the TOBY app</li> <li>• Novelty wore off</li> <li>• Was not a game, and did not have music, so the child was not motivated</li> <li>• Child perseverated on the rewards and not the activities</li> <li>• Difficulty staying engaged after the favourite activities were completed.</li> <li>• Got bored towards the end of the trial</li> <li>• Child got frustrated because they wanted other apps on the iPad, and as there were no others, would not use the iPad.</li> </ul>

Topic	Themes	Common responses
	Better for the earlier stages of development	<ul style="list-style-type: none"> <li>• Enjoyed using it when child was younger</li> <li>• Too easy and not benefiting the child</li> <li>• Parent does not see point in using it, as it will be no benefit due to tasks being too easy now.</li> </ul>
	Challenge finding the “just right” level for child	<ul style="list-style-type: none"> <li>• Could not skip levels so did not have time to get to the activities that would be beneficial for the child.</li> </ul>
Skill Maintenance	Maintained skills	<ul style="list-style-type: none"> <li>• 12 out of 15 parents reported maintenance in at least one skill area in language, daily living skills and social communication, such as listening to and following instructions</li> </ul>

## 6.5 Discussion

Outcomes of this study follow on from the findings by Parsons et al. (2019) who reported limited effectiveness of the TOBY app in improving the receptive language, social and symbolic communication, pragmatic language when analysing pre-, post- and follow-up data of children with ASD. When the findings from the study by Parsons et al. (2019) and this longer-term follow-up study are combined, it can be concluded that participants who use the TOBY app for three months gained and maintained skills for up to at least 12 months for receptive language, social and symbolic communication, pragmatic language and playfulness. However, due to the absence of a control group to account for confounding variables, such as natural development and a smaller sample size at the 12-month follow-up, caution should be exercised when interpreting the findings. No skill improvements were detected in the SPT and the MSEL visual reception, fine motor and expressive language subscales between pre- and post-intervention.

This study is one of a few to investigate the long-term outcomes of a parent-mediated ICT-based psychosocial intervention for children with ASD (Estes et al., 2015; Pickles et al.,

2016). These findings extend the current knowledge about parent-mediated ICT-delivered interventions for children with ASD, indicating the limited effectiveness of the TOBY app for children with ASD who live in regional areas. Together with findings from Pickles et al. (2016) and Estes et al. (2015), results from this study contribute to the emerging evidence to support the long-term benefits and maintenance of skills where parents are active agents in the delivery of therapy for children with ASD who live in regional areas.

The TOBY app was anticipated to lead to improvements in the longer term for the skills of language, social communication and playfulness as the children developed, given the TOBY app's focus on fundamental skill development in these areas. That is, the TOBY app curriculum includes tasks targeting skills in early child development, which can be built on and generalised to more complex skills as the child develops (Moore et al., 2015; Venkatesh et al., 2013). Interestingly, only skills in social communication, as measured by the CSBS, continued to improve significantly after the cessation of the intervention, while no continued improvements were noted for the other outcome measures. A possible explanation for this finding could be a ceiling effect of the MSEL instrument, resulting in a decreased sensitivity to detect change as children approach the upper end of possible scoring. However, the POM and ToP outcome measures have demonstrated psychometric validity and reliability for older children, so have higher responsiveness to detect development in these skills for participants in this study.

Another plausible explanation for this finding is the progressive attenuation of treatment effect from the child's interaction with the parent to the child's interaction with the assessor, similar to the pattern observed in the Preschool Autism Communication Trial (PACT) RCT conducted by Green et al. (2010). Further, the findings in this study are congruent with the findings reported in the PACT RCT, whereby parent-mediated



interventions positively impacted the parent-child dyadic social communication in the long-term, as measured by the parent-rated CSBS in this study. Despite no statistically significant improvements the MSEL, POM and ToP outcomes from T2 to T3, the overall findings across all measures did not detect a decline in any skills over the follow-up period, supporting the maintenance of these skills.

In the RCT from which the participants were recruited, pre-post intervention analysis of the pooled participant data (n=57) showed changes in the sub-scale of receptive language in the MSEL, social and symbolic subdomains of the CSBS, and the POM but not playfulness (Parsons et al., 2019). Interestingly, in this study when follow-up was extended to 12 months or greater, the playfulness of the children did improve significantly over the intervention period (T1 to T2). However, there was no statistically significant improvement from T2 to T3 in the ToP, suggesting no developmental gains in play over this time period.

While playfulness was not a targeted skill area within the TOBY app curriculum and was not a primary outcome in this study, skills such as receptive and expressive language, joint attention, and gestures learnt from the TOBY app could be vital precursors in the development of children's playfulness (Kasari, Gulsrud, Freeman, Paparella, & Helleman, 2012; Moore et al., 2015). Playfulness, as a construct measured by the ToP, is determined by evaluating the presence of internal control, intrinsic motivation, the freedom to suspend reality, and skills related to framing (Bundy, 2004; Cordier, Bundy, Hocking, & Einfeld, 2009). Relevant to the skills the TOBY app targets, framing in the construct of playfulness requires skills in the ability to read and give social cues (Bundy, 2004). It was anticipated that the language and social communication skills learnt during the trial period could have resulted in improvements in the playfulness of the child; however, these were not detected at 12 months follow-up. This could indicate that generalisation of social communication

skills taught in the TOBY app to spontaneous play interactions are limited, reinforcing similar findings in play-based interventions and highlighting the need for targeted interventions for children with ASD to improve skills in play (Henning, Cordier, Wilkes-Gillan, & Falkmer, 2016; Ingersoll & Schreibman, 2006; Williams White, Keonig, & Scahill, 2007).

Twelve out of 15 parents reported their child had maintained at least one skill at 12 months post-intervention in one of the areas of receptive language, social communication or daily living skills, despite them no longer using the TOBY app. Triangulation by analysing the individual changes for each child confirmed these parent reports, thus, supporting the findings of skill maintenance in this study.

Further, responses from the parents indicated 13 out of the 15 children were no longer using the TOBY app after twelve months, citing a lack of time and a loss of interest from their child as common reasons. The cessation of the TOBY app after three-months was mirrored in the study by Whitehouse et al. (2017), who reported a significant decline in per day median use from approximately 19 minutes during the first three months, to a per day median use of two minutes over the following three months. Parents from the Whitehouse et al. (2017) reported the main reasons for discontinued use were the same as reported in this study, a lack of time and loss of interest in the TOBY app by their child. The discontinued use of the TOBY app could be viewed from a positive perspective. That is, participants may have ceased the TOBY app due to the child having developed the skills the TOBY app was targeting and maintained the skills; therefore, parents no longer felt the need to use the TOBY app anymore. Parent responses support this explanation with many reporting the TOBY app was too easy and more suitable for younger children, with their children having surpassed the level taught in the TOBY app.

### **6.5.1 Implications for future research and clinical application of ICT-based interventions**

The development and evaluation of ICT-based interventions for more complex skills in older children with ASD are warranted. The key reasons as reported by the parents for the cessation of the intervention was their child losing interest in using the app and the tasks became too easy for the child. When developing the curriculum, developers and researchers should consider the scope of the curriculum and could include tasks and activities that target higher-level skills and are appropriate for children who are developmentally more advanced.

Larger scale studies, using control groups with an a priori long-term data collection point in the study design could improve the generalisation of results to broader populations, which was a limitation of this study. Lastly, researchers should focus on the most effective components of ICT-based interventions to improve the generalisability of skills learnt through ICT-based intervention, a common limitation of cited in the literature of ICT-based interventions for children with ASD (Grynszpan, Weiss, Perez-Diaz, & Gal, 2014; Silver & Oakes, 2001).

### **6.5.2 Limitations**

A number of limitations are present in this study. Consideration of the findings in the context of the study design is required. The study followed-up with a small non-randomised sample, limiting the ability to control for confounding factors. Further, given the lack of a control group, which was unavoidable due to ethical considerations, the development of skills in the children with ASD could be attributed to typical development in the children or the treatment-as-usual they received during the study. However, the lack of statistically significant improvements from T2 to T3 in the MSEL, POM and ToP suggest this may not necessarily be the case. As this was a long-term follow-up, the age of

the participants resulted in a ceiling effect in some of the outcome measures as the instruments were chosen to assess younger, less developed participants in the RCT effectiveness trial. Specifically, a number of participants achieved ceiling scores in the CSBS speech (40%) and SPT (33%) outcome measures, thus, decreasing the sensitivity of these measures to detect change; however, given no statistical difference was detected, the effect on measuring maintenance of skills is still valid for these outcome measures.

## **6.6 Conclusion**

This study investigated the long-term follow of an ICT-based intervention, the TOBY app, for children with ASD living in regional Australia to determine the maintenance or continued improvement their language and social communication skills after at least 12 months post-intervention. Findings demonstrate the receptive language, social skills, pragmatic language and playfulness of children with ASD improved during the three-month intervention period and were maintained at least 12 months after ceasing the TOBY app intervention.

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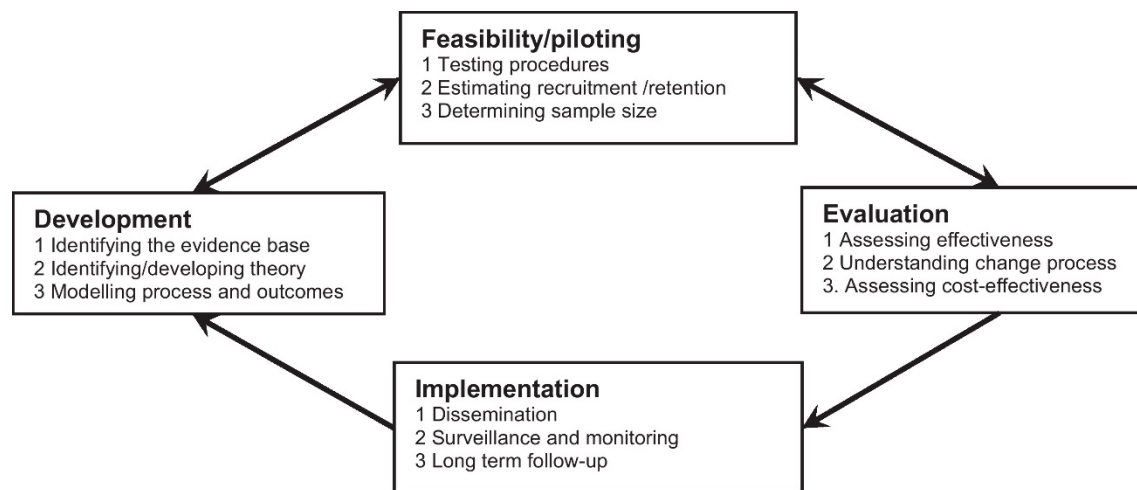
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## Chapter 7 Discussion and Conclusion

*In order for efficacious interventions to be successfully implemented, the community context must be considered explicitly throughout all phases of research* (Dingfelder & Mandell, 2011, p. 603).

### 7.1 Overview

The main focus of my research was to evaluate an information communication technology (ICT)-based intervention the Therapy Outcomes By You application (TOBY app) in the regional and remote Western Australian (WA) context. In this chapter, I integrate the findings from each research phase to answer the core research question — is the TOBY app a feasible, effective and appropriate intervention? I use the United Kingdom Medical Research Council guidelines for developing and evaluating complex interventions (referred to as the UKMRC guidelines henceforth) to structure my findings (Figure 7.1). Next, I discuss the implications of these findings for researchers working with families of children with ASD living in regional WA. Following this, I present recommendations for incorporating the Diffusion of Innovation theory with the UKMRC guidelines for the development and evaluation of ICT-based interventions. I then highlight areas of future research for ICT-based interventions. Finally, I acknowledge the limitations of research before finishing with my conclusions.



**Figure 7.1 Design and Evaluation of Complex Interventions (Craig et al., 2013)**

## **7.2 TOBY app as a feasible, effective and appropriate intervention**

### **7.2.1 Development phase**

Guided by the development stage of the UKMRC guidelines, I reviewed existing scientific evidence on innovative models of service delivery for families of children with ASD, living in regional and remote areas (Craig et al., 2013). Two systematic reviews had been undertaken to investigate the effectiveness of ICT-based interventions for children with ASD (Ramdoss et al., 2011; Ramdoss et al., 2012). The studies reported that ICT-based interventions demonstrated statistically significant improvements in communication, social and emotional skills. However, due to the small effect size detected across the existing studies and a lack of high-quality studies (i.e., poor study design, low participant numbers), ICT-based interventions should not yet be considered as an evidence-based approach to teach communication, social and emotional skills to children with ASD.

Given the limited literature investigating the effectiveness of ICT-based interventions for families of children with ASD living in regional and remote areas, I expanded the terms of my literature search to include ‘parent-mediated interventions’. This decision was guided by the philosophical underpinning of ‘*teaching the parent how to teach*’ to provide parents

with skills to deliver evidence-based Applied Behaviour Analysis (ABA) therapy during the crucial period following diagnosis, which often coincides with no access to formal therapy (Venkatesh, Phung, Duong, Greenhill, & Adams, 2013). This led to the decision of reviewing existing literature on parent-mediated interventions as the mode of intervention delivery. In their Cochrane review, Oono, Honey, and McConachie (2013) concluded that some evidence existed for the effectiveness of parent-mediated interventions, with stronger evidence supporting improvements in parent-child interactions and weaker evidence supporting improvements in child language comprehension and ASD symptom severity. These scoping exercises led to the conclusion that there was an emerging research area investigating the use of parent-mediated interventions delivered remotely for families and their children with ASD living in regional and remote areas; however, a systematic review on these interventions had not been completed. Hence, I undertook a systematic review (Chapter 2) to investigate parent-mediated interventions delivered remotely for families of children with ASD living outside of major cities.

Five electronic databases (CINAHL, Embase, ERIC, PsycINFO, and Pubmed) were searched for studies investigating parent-mediated interventions for families living outside of major cities who have a child with ASD. Seven studies met eligibility criteria including two randomised controlled trials, three multiple-baseline studies, and two pre-post cohort studies. The populations from the included studies resided in either the United States of America, Canada or Australia. Parent-mediated interventions included self-guided websites, training videos and video-conferencing. Key findings from this systematic review were: 1) web-based delivery methods, DVDs and telehealth all had some effect in improving treatment fidelity and increasing parents' knowledge in ASD; 2) intervention training for parents delivered using video were more effective and accepted by parents compared with written training; and 3) regular contact with therapists resulted in improved

program completion, intervention fidelity, parent engagement, intervention appropriateness, and parent's positive perception of their children when compared to self-directed (i.e., minimal therapist contact) programs.

Chapter 2 highlighted several limitations that compromised the quality of the studies in this area of research, including: 1) lack of rigour in study designs; 2) infrequent use of standardised outcome measures; 3) a lack of measurement of child outcomes; 4) small sample sizes; and 5) high risks of bias. Due to the heterogeneity between samples and the variability in classification systems and definitions adopted by the authors when describing participants from regional and remote areas, it is challenging to generalise the findings to specific populations. As a result, the ability of the reader to make informed interpretations from the research were diluted.

The key recommendations for future research from Chapter 2 were: 1) further investigation into the unique context of families of children with ASD living in regional and remote settings is required; 2) researchers should employ rigorous methodologies to improve the quality of the evidence for parent-mediated interventions delivery remotely, such as using randomised controlled trials (RCT) and standardised outcome measures; 3) researchers should aim to use standard geographical classification systems when describing their participants living in regional and remote areas to improve the applicability and generalisation of findings for the reader; and 4) further investigation into parents' perceptions of the appropriateness of parent-mediated interventions is needed. In conjunction with existing literature for ICT-based interventions, the recommendations from Chapter 2 informed the next phases of my research.

Chapter 3 addressed recommendation one from the systematic review, which was to investigate the unique context of families of children with ASD living outside of a densely

populated area. This led to the development and implementation of a cross-sectional survey for parents of children with ASD living in WA (Chapter 3). For this study, there was a focus on recruiting families from regional and remote WA, with the aim to compare responses of parents who live in more densely populated areas to those who live in less densely populated areas. A secondary aim of this study was to compare the stress, coping, QoL, and daily routines of all parents of children with ASD living in WA to the general population using Australian normative data.

Recommendations two and three from Chapter 2 led to and guided the development and implementation of an RCT described in Chapter 4 to investigate the effectiveness of the TOBY app. To this effect I incorporated the following recommendations: 1) including standardised child outcomes measures; 2) using the Australian Standard Geographical Classification (ASGC) to describe participants; 3) aiming to recruit the required participant numbers to execute an appropriately powered RCT; and 4) using an RCT to minimise selection bias. Recommendation four from the review informed Chapter 5, which was an investigation into the parent experiences of using the TOBY app to assess the appropriateness of the intervention.

### **7.2.2 Feasibility and piloting phase**

A feasibility/piloting study of the TOBY app, as per the UKMRC guidelines, had been previously conducted by the developers prior to the commencement of my research (Moore et al., 2015; Venkatesh, Greenhill, Phung, Adams, & Duong, 2012; Venkatesh et al., 2013). The evaluation of complex interventions is often undermined by problems of appropriateness, adherence, poor fidelity in the delivery of the intervention, poor recruitment and retention, and smaller than expected effect sizes (Craig et al., 2013; Eldridge, Ashby, Feder, Rudnicka, & Ukoumunne, 2004). In acknowledgement of the



importance of factors related to the feasibility of an intervention, the UKMRC guidelines dedicate an entire stage to the process of testing an intervention's feasibility.

The pilot trial by Moore et al. (2015, p. 217) concluded that the TOBY app might make a useful contribution to early intensive behavioural intervention (EIBI) for children with ASD by delivering “high rates of appropriate learning opportunities”. Moreover, the authors reported that even in the absence of therapist support, families were able to utilise the TOBY app easily and extensively. The pilot paper did have some notable limitations. Firstly, a key aspect in the development phase that was absent was the evaluation of the intervention's appropriateness — a vital component in evaluating an intervention (Craig et al., 2013; Evans, 2003). Secondly, there were no pre-post outcome measures collected to assess the intervention effects on the skill areas the TOBY app were targeting. Thirdly, the pilot trial lacked a control group for comparison, hence, controlling for confounding variables was not possible. Finally, the sample population were from a major city. As such, the findings were limited in their generalisation to families living in regional and remote areas given their unique social context.

Prior to any further evaluation of the TOBY app, I wanted to investigate whether the geographical location of families of children with ASD living in regional and remote WA influenced their stress levels, coping strategies, QoL and daily routines, given the access barriers experienced by families living in less densely populated areas (Dew et al., 2013; Dew et al., 2012). These family contextual factors of stress, coping strategies, QoL, and daily routines may impact the feasibility of the TOBY app for families living in regional and remote areas. Firstly, I hypothesised that increased stress and disrupted daily routines would negatively impact parents' capacity to deliver the intervention, given the relatively high demand in adhering to the prescribed dosage of the TOBY app (i.e., 20 minutes per

day). Secondly, findings from this study could provide useful insights for the TOBY app developers, and possibly other ICT-based intervention developers, to be better informed to tailor ICT-based interventions to the local context in which the intervention was being implemented, an essential aspect of developing and evaluating a complex interventions (Craig et al., 2013). Lastly, findings from this phase of the research would expand the current knowledge base of parents with children with ASD living in regional and remote WA, which could help inform service providers and policymakers to address the factors that serve as barriers in the provision of support services to this population.

In Chapter 3, I sought to compare the coping, stress, QoL and daily routines of parents of children aged two to 18 years with ASD living in low densely populated (LDP) areas to those parents of children aged two to 18 years with ASD living in densely populated (DP) areas in WA. A cross-sectional survey was administered to 278 parents of children with ASD using validated self-report measurements on stress, coping strategies, QoL and daily routines. I used the ASGC (informed by Chapter 2, Recommendation 3) to classify the sample into LDP areas comprising of families living in outer regional, remote and very remote locations, with DP areas consisting of parents living in major cities and inner regional areas. This grouping was based on the expected similarities in service access of each of the geographical classifications, that is, I hypothesised that parents living in major cities and inner regional areas would experience similar travel distances to access services (Australian Bureau of Statistics, 2011).

Key findings from Chapter 3 were: 1) parents from LDP areas were more likely to adopt avoidant coping strategies compared to parents living in more DP areas, albeit with a small effect size ( $p = 0.004$ ;  $Exp [\beta] = 1.14$ ); and 2) when both groups (parents living in DP areas and in LDP areas) were combined, parents of children with ASD experienced higher

levels of stress and poorer QoL compared to normative population data in Australia. Aside from these findings, there appeared to be little difference between the populations in relation to coping strategies, levels of stress, QoL and daily routines.

Overall, these findings suggest that while having children with ASD presents parents with some challenges of increased stress and poorer QoL, which is consistent with published literature (Hayes & Watson, 2013; Vasilopoulou & Nisbet, 2016), their geographical location does not seem to have a significant impact on the parents' coping strategies, stress, QoL or daily routine. Given the higher levels of stress and poorer quality of life experienced when compared to the broader population, there is still merit in ensuring parents of children with ASD can access feasible, effective and appropriate interventions for their children. Ensuring access to these interventions may ameliorate some of the stress and impact on QoL experienced by this population, potentially resulting from their children's behaviours. In their pilot paper, Moore et al. (2015) reported broad patterns of use of the TOBY app, citing contextual issues such as high levels of parental stress as a possible reason for non-use of the TOBY app. As little difference was detected between parents living in DP areas to living in LDP areas for parents of children with ASD, it is plausible to infer that parents living in LDP areas should be equally equipped to deliver ICT-based interventions when compared to their DP counterparts.

When combined with the pilot trial by Moore et al. (2015), these findings provide emerging evidence supporting the feasibility of using the TOBY app. With few differences between the coping, stress, QoL and daily routines between parents living in LDP areas and those in DP areas, the TOBY app could be a feasible intervention for all children with ASD and their families, regardless of where they live. There may, however, be other factors unique to living in regional and remote locations that influence the feasibility, effectiveness

and appropriateness of the TOBY app. Thus, further research with this population is warranted. Together, the findings from Chapter 2, Chapter 3, and the feasibility study by Moore et al. (2015) supported the progression from the feasibility stage of the UKMRC guidelines to the evaluation phase. Therefore, the RCT as outlined in Chapter 4 was undertaken.

### **7.2.3 Evaluation phase**

#### **7.2.3.1 Randomised controlled trial**

In Chapter 4, I conducted an RCT with 59 participants demonstrating that children with ASD who used the TOBY app for three months, at a prescribed dosage of 20 minutes per day, experienced significant improvements in expressive language when compared with the control group of children with a developmental age between two and six years. Further, as all participants in the study (both control and intervention group) received the intervention due to the waitlist study design, when all of the participants were pooled and measured pre-post intervention as a single cohort ( $N = 59$ ), significant improvements were detected in receptive language, pragmatic language and social communication skills (symbolic and social). It should be noted that the study did have some limitations, namely the poor adherence to the prescribed dosage of 20 minutes of therapy per day (mean = 11 minutes per day), a notable drop-out rate (15%) of participants, and lack of data on the nature and quantity of participants' treatment-as-usual. The absence of a control group to ameliorate confounding variables for the analysis may limit the veracity of this finding as these changes could be as a result of natural development. However, the post-hoc analysis and larger sample achieved through pooling participants indicate improvements in these skill areas following use of the TOBY app.

As discussed in Chapter 1, in parallel with my study, Whitehouse et al. (2017) conducted a

similar trial using a multi-centre RCT to evaluate the effectiveness of the TOBY app with children aged two to six, with participants primarily living in major cities. Importantly, while we were investigating the same intervention with similarly aged children, the location of the families was distinct. In the Whitehouse et al. (2017) study, 80 participants were recruited and randomised into a therapy-as-usual group ( $n = 39$ ) and a therapy-as-usual group plus 20 minutes per day of the TOBY app ( $n = 41$ ) over six months.

Participants who completed the TOBY app intervention and therapy-as-usual demonstrated statistically significant improvements in visual motor and fine motor skills when compared to the control group. Interestingly, the study did not report any improvements in expressive language. Poor adherence was noted in the intervention group, with usage dropping on average from 19 minutes per day over the first three months to two minutes per day over the final three months. Importantly, in their trial, Whitehouse et al. (2017) also experienced a notable participant drop-out rate (23.1%) in their treatment group, which was slightly higher than what was recorded in my RCT (15%; Chapter 4).

In summary, when the results from the studies are combined, it can be concluded that the TOBY app demonstrates a weak effect to positively influence children with ASD's skills in receptive and expressive language, visual motor skills, fine motor skills, social communication skills, and pragmatic language. As an outcome of Chapter 4, a recommendation for future research was the need to investigate the lived experience of using the TOBY app from the perspective of the parents. Specifically, further research was required to investigate the appropriateness of the intervention. This outcome was congruent with an a priori decision to conduct interviews exploring parental experiences to investigate the TOBY app's appropriateness in accordance with the UKMRC guidelines, given this had not been undertaken by the developers in their pilot trial (Moore et al., 2015).

### 7.2.3.2 Appropriateness

In Chapter 5, I conducted interviews with 24 parents of children who participated in the RCT (Chapter 4), with the aim of capturing their experiences of using the TOBY app. From these interviews, themes were generated using the thematic analysis approach by Braun and Clarke (2006). Following the thematic analysis, I applied a theoretical model to these themes to evaluate the appropriateness of the intervention from the perspectives of the parents. Findings from Chapter 5 partially supported the TOBY app as an appropriate intervention for children with ASD living in regional and remote WA.

The thematic analysis generated one core theme and two major themes. The core theme was, *The TOBY app was not a panacea* for all their children's barriers. Major themes identified were: 1) *if you have met one child with Autism, you have met one child with Autism*, indicating that the TOBY app was more appropriate for some children with ASD, but less so for others; and 2) *the TOBY app is just one piece of the jigsaw*, meaning parents suggested that the TOBY app should be just one part of the therapy landscape, and cannot and should not replace face-to-face therapy, but complement it.

When the identified themes were mapped against an appropriateness framework, the key findings from Chapter 5 were that collectively, parents who participated in both the RCT (Chapter 4) and interviews (Chapter 5) felt the TOBY app was relevant and important to them and their children's needs. They expressed partial support for using the TOBY app as: 1) a positive experience for them and their children; 2) beneficial for them and their children; 3) a socially and ecological valid intervention; and 4) an intervention that supported change and continuation in the skills learnt.

The final stage of my research progressed to the implementation stage of the UKMRC guidelines. In this stage, a longer-term follow-up study was conducted to determine if a

sample of participants from the RCT maintained their gains for at least 12 months post-intervention.

## **7.2.4 Implementation Phase**

### **7.2.4.1 Long-term follow-up**

In Chapter 6, to determine if participants maintained or continued to improve their language and social communication skills after at least 12 months post-intervention, I completed a long-term follow-up with 15 children who participated in the RCT. The UKMRC guidelines recommend that long-term follow-up, while uncommon, is very important as it informs whether short-term changes detected in effectiveness studies were maintained. Furthermore, the study also aimed to explain the maintenance of skills and ongoing use of the TOBY app from the perspectives of the parents who participated in the RCT.

Findings from Chapters 4 and 6 suggested that participants who use the TOBY app for three months maintained gains for 12 months in receptive language, social and symbolic communication, pragmatic language and playfulness. That is, there were significant improvements between pre- and post-intervention, with no significant decline detected between post-intervention and follow-up after 12 months. No skill improvements were detected in participants' symbolic play, visual motor, fine motor and expressive language skills between pre- and post-intervention.

Most families (13 out of the 15) were no longer using the TOBY app after twelve months, with a lack of time and a loss of interest in the TOBY app from their child as the main reasons provided. The discontinued use of the TOBY app could be viewed as a positive outcome for the child and their parents. That is, families may have ceased using the TOBY app in response to their child having attained the skills targeted by the TOBY app and,

subsequently, maintained those skills, thus the parents decided they no longer needed to use the TOBY app. Responses from parents in Chapter 6 support this explanation with many parents reporting that the TOBY app became too easy and the activities were more suitable for younger children.

### **7.2.5 Summary**

In summary, the UKMRC guidelines for the development and evaluation of complex interventions steered my evaluation of the TOBY app as a parent-mediated ICT-based intervention for children with ASD with a developmental age of two to six who live in regional and remote WA. From my research, it can be concluded that parents of children with ASD can deliver interventions to their children in their own homes and that parents living in regional and remote areas experience no greater barriers due to stress levels, QoL, coping mechanisms and daily routines to deliver these interventions when compared to their major city-dwelling counterparts. The TOBY app was subsequently evaluated using an RCT. Only one significant improvement was detected in the RCT between the control and intervention groups (expressive language), with significant improvements detected in receptive language, pragmatic language and social communication skills (symbolic and social) when participants were pooled and measured pre-post intervention as a single cohort. The RCT had a notable drop-out rate and treatment fidelity showing poor adherence to the prescribed dosage. Furthermore, findings from Chapter 5 demonstrated that parents only partially supported the TOBY app as an appropriate intervention for their children with ASD.

Overall, the results from this research provide partial support for the TOBY app as a feasible, effective and appropriate intervention for families of children with a developmental age of two to six years with ASD living in regional and remote WA. The



findings from my research do, however, provide some implications and clear directions for future research and some learnings that would be valuable in the development of future ICT-based interventions for children with ASD.

### **7.3 Implications for research and the development of ICT-based interventions**

#### **7.3.1 Importance of consistency when describing participants from regional and remote areas to improve the generalisation of research results**

One of the biggest challenges I faced in the initial stages of my research and in generalising its findings was the inconsistencies in definitions and the operationalisation of regionality in the literature. The lack of specificity by researchers when describing regional and remote participants was highlighted in Chapter 2. A strong recommendation I have for future researchers working with people from regional and remote areas is to use standardised geographical classification frameworks to aid the interpretation of findings. If standard geographical classification frameworks are not available, then researchers should provide detailed demographic and geographical data, such as distance to major cities and available health infrastructure, to improve the interpretation of findings. The lack of defined geographical features provided in studies where participants lived in regional and remote areas made it challenging when I tried to link and apply my research findings to the current literature. In their work, Smith, Humphreys, and Wilson (2008) highlighted challenges when defining geographical location, and commented on the trend in the literature for researchers to use terminology, criteria and qualifiers that were accepted as meaningful to them in the context of their work, regardless of the limitations on generalisations that the readers can derive from their findings.

Researchers working with families who live in regional and remote areas should not assume that the characteristics and experiences of families within their sample are the same

as all other families living in regional and remote areas. Indeed, in the Australian context, the discrepancy in lived experience between families living in inner regional centres, compared with those living in very remote areas can be starkly different (Dew et al., 2012; Smith et al., 2008). To group families living in regional and remote areas as having the same characteristics as the major city dwelling counterparts would be disingenuous and limits the finding's generalisability to the broader population. Finally, when research methods dictate that grouping of participants from different regional classifications must be done, researchers should provide a strong rationale for their groupings.

### **7.3.2 Innovative Delivery Models for ICT-based Interventions**

A potential model that could address the deficits of the TOBY app that still combats the access barriers often experienced by families of children with ASD is the use of telehealth in combination with the TOBY app (Heitzman-Powell, Buzhardt, Rusinko, & Miller, 2014; Lincoln et al., 2014; Wainer & Ingersoll, 2015). A key finding in Chapter 5 was the parents' desire to receive ongoing support when implementing the TOBY app. Face-to-face intervention and support to parents could be delivered using video conferencing; an established modality used to provide training and support to parents implementing interventions, in combination with the TOBY app, thus limiting the travel time for families living considerable distances from services (Boisvert, Lang, Andrianopoulos, & Boscardin, 2010; Pickard, Wainer, Bailey, & Ingersoll, 2016; Vismara, Young, & Rogers, 2012; Wainer & Ingersoll, 2015).

A blended model combining both telehealth and ICT-based interventions holds promise given the preliminary evidence supporting the efficacy of each intervention by itself (Boisvert et al., 2010; Ramdoss et al., 2012). Findings from Chapters 2 and 5 provide some guidance to clinicians for the types of factors that should be considered when incorporating

ICT-based interventions delivered remotely into their treatment plans.

Future research should focus on estimating the correct dosage of face-to-face contact via video conferencing and use of the TOBY app. Furthermore, there are calls for other innovative service delivery methods that incorporate ICT in their delivery methods but are augmented with other supports when sufficiently trained health professionals are lacking. The model could include the training of education assistants, disability support workers or other staff delivering evidence-based ASD interventions in conjunction with ICT-based interventions. A mixed model like this could be more feasible and appropriate in the delivery of services to families living in regional areas when compared to more traditional face-to-face models of service delivery (Dew et al., 2013; Dew et al., 2012).

Worth noting is that innovative service delivery models are not solely for families living in regional and remote areas. Given the known burden (financial, psychosocial) on all families of children with ASD to access therapy services, innovative ICT-based interventions may be appropriate for families living in major cities, thereby improving the usage of EIBI interventions. Supporting parents to more easily access interventions for their children could contribute in ameliorating the stress and poorer QoL that parents of children with ASD experience in WA, compared to the general population reported in Chapter 3.

### **7.3.3            Integration of Diffusion of Innovation theory with the UK framework to improve the design of ICT-based interventions**

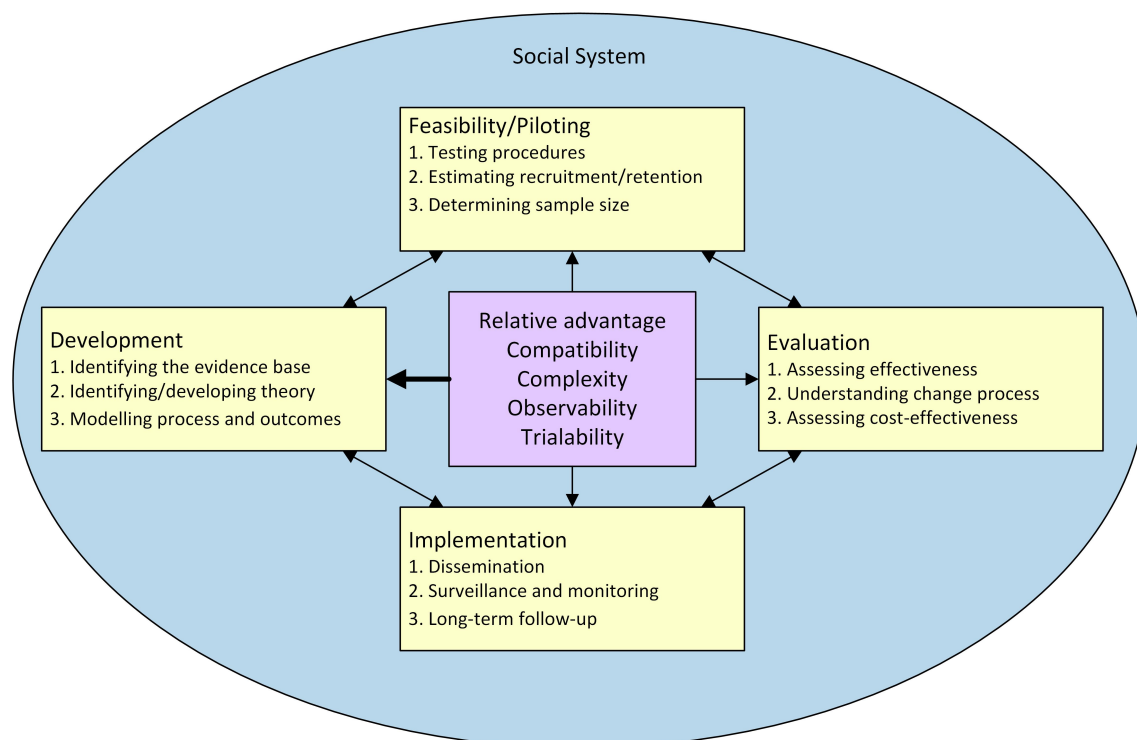
Despite families voluntarily electing to participate in the trial and the provision of a free intervention and iPad, one of the biggest challenges faced in the RCTs of the TOBY app [both mine and the Whitehouse et al. (2017) study] was poor treatment adherence and notable drop-out rates. These findings are indicative that the TOBY app may have some

issues with the adoption of the technology. Accordingly, I am of the opinion there is value in trying to answer the question, “What needs to be done differently to increase adherence and continued use of ICT-based interventions?”

To answer this question, developers and researchers of ICT-based interventions should consider how their interventions will be adopted by the end-user; to not only improve intervention fidelity, but ensure widespread use within the ASD community. The Diffusion of Innovation theory and the innovation-decision process, a key aspect of that theory, were introduced in Chapter 1 (Rogers, 2003). While the UKMRC guidelines and appropriateness literature aided in guiding the methodology for the evaluation the TOBY app, they are not well suited in explaining the drop-out rate and poor adherence in the RCTs, or in providing recommendations on how to minimise these factors in future studies. Consideration of the Diffusion of Innovation theory can aid developers and researchers of interventions for children with ASD to design interventions that increase the likelihood of the intended client group adopting their interventions. In isolation, the three approaches (UKMRC guidelines, appropriateness and Diffusion of Innovation) serve different purposes; however, in combination, I believe they may help more families of children with ASD to adopt evidence-based ICT-based interventions.

In the section that follows, I propose that researchers and developers of ICT-based interventions who use the UKMRC guidelines could integrate aspects of the Diffusion of Innovation theory within their design and evaluation process to increase the likelihood of adoption and diffusion throughout the ASD community. A schema demonstrating how this could be operationalised is outlined in Figure 7.2. Firstly, I will discuss the importance of the social context in relation to ICT-based interventions and how unique aspects of the Diffusion of Innovation theory can be applied to social systems. Secondly, I will provide

an overview of how consideration of the characteristics of the interventions are vital in the innovation-decision process and suggest how these characteristics can be integrated into the UKMRC guidelines. Thirdly, I discuss how an aspect of the Diffusion of Innovation theory, known as *preventive innovations*, could provide a unique perspective for researchers and developers. Lastly, I will provide recommendations for each stage of the UKMRC guidelines for researchers and developers to utilise the integrated model to improve the likelihood of adoption of innovative, evidence-based interventions for children with ASD.



**Figure 7.2 Integration of Diffusion of Innovation and the UKMRC framework (Craig et al., 2013; Rogers, 2003)**

### 7.3.3.1 Social System

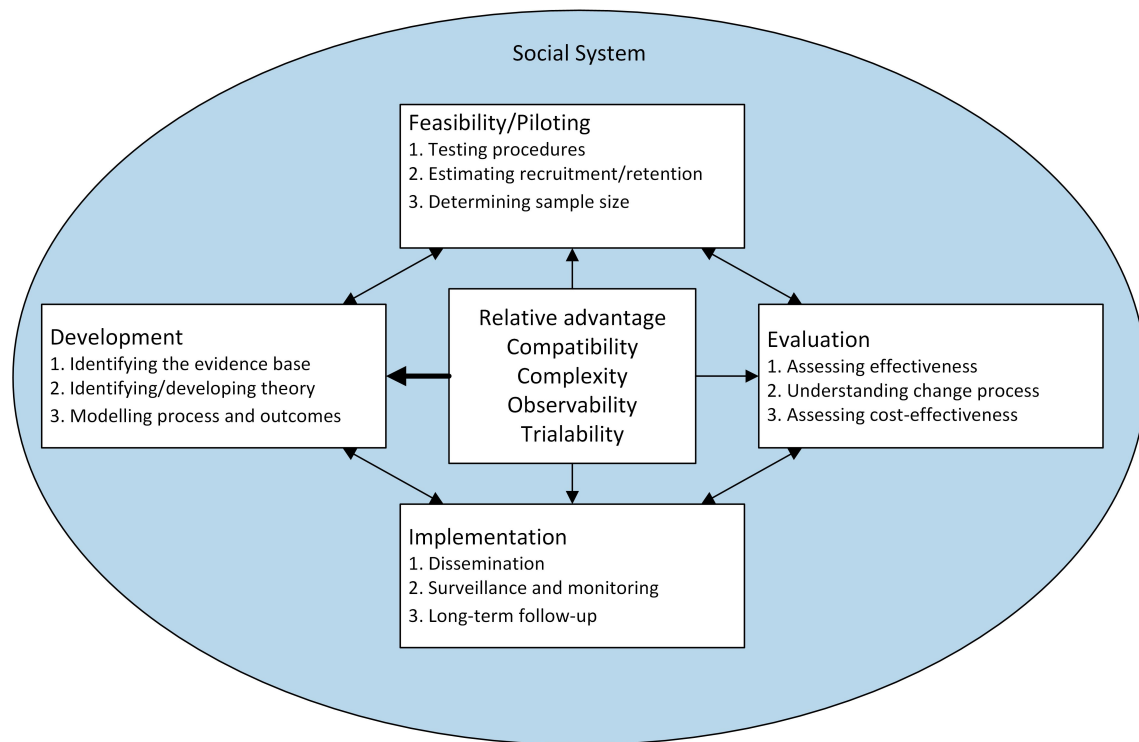
I propose that consideration of the social systems should be given in all stages of the design and evaluation process of interventions to improve the likelihood of adoption. While the authors of the UKMRC guidelines encourage intervention developers to consider the social

context within which an intervention is implemented and evaluated, in isolation, the UKMRC guidelines place a larger emphasis on the social context during the development and evaluation phase, with less emphasis on the other phases. By integrating the Diffusion of Innovation theory with the UKMRC guidelines, the social context receives greater importance throughout the entire process; not just in the development and evaluation phases (Campbell et al., 2007; Craig et al., 2013; Rogers, 2003). Moreover, the Diffusion of Innovation provides researchers and developers with some clear aspects (i.e., felt needs, social norms, previous behaviours) of the social context that improve the chance of adoption that are not explicitly identified in the UKMRC guidelines.

Key aspects of the social context that Rogers (2003) suggest will influence parents' decision-making can be drawn from the innovation-decision process discussed in Chapter 1. Specifically, Rogers (2003) reports that prior to starting the innovation-decision process, the norms of the social system within which the intervention is going to be implemented need to be well understood. Examples of these norms are consideration of the community's technology literacy, the expectations within the social system in relation to caregiver roles (e.g., parents not wanting to be their children's therapist as well as their parent), and the false assumption that all children with ASD have an affinity for ICT. Further, characteristics of the social system such as socioeconomic characteristics and communication behaviours will also influence the decision-making process. These need to be considered at all stages in the design and evaluation stages, and not just in the early stages.

In relation to regional and remote families of children with ASD, there is much to be gained by developers and researchers fostering an improved understanding of the unique social systems in which these families live compared to their major city counterparts.

Understanding the unique social contexts of regional and remote families of children with ASD could improve the successful adoption of ICT-based intervention at all stages. This integration is represented in the framework with the coloured circle that encompasses all phases of the UKMRC guidelines (see Figure 7.3).



**Figure 7.3 Social context in the UKMRC framework**

Findings from Chapters 2 and 5 support the need for researchers and developers to better understand the social context, suggesting that the limited support provided in the RCT could have contributed to the reduced adherence and a high drop-out rate of participants in the study. Further, data from Chapter 5 highlighted the need for ongoing support for parents from therapists when using the app, despite the acknowledgement that the TOBY app was easy to use and provided some benefits for children with ASD. Although parents rated the complexity of the TOBY app as low, a characteristic likely to improve adoption, the higher drop-out rate and poor adherence to the recommended dosage could be due to a failure to fully account for the intricacies of the social context during the evaluation stage.

The importance of the social system in relation to the Diffusion of Innovation theory is not new to research in ASD interventions. In their paper, Dingfelder and Mandell (2011) applied the Diffusion of Innovation theory to understand why effective innovations (interventions) for children with ASD are not successfully adopted or implemented in the public health or education systems. A key finding from this paper supports my recommendation for the need for intervention developers to consider the real-world context in which the intervention is being implemented in parallel to intervention development. Moreover, the authors stress that developers and researchers should: 1) partner with community partners and organisations to facilitate the successful adoption, implementation and maintenance of developed interventions; and 2) develop new interventions in collaboration with communities to ensure they meet the needs and demands of that community, thus improving the likelihood of successful adoption. Strategies on how these findings could be operationalised are discussed in the recommendations section of this chapter.

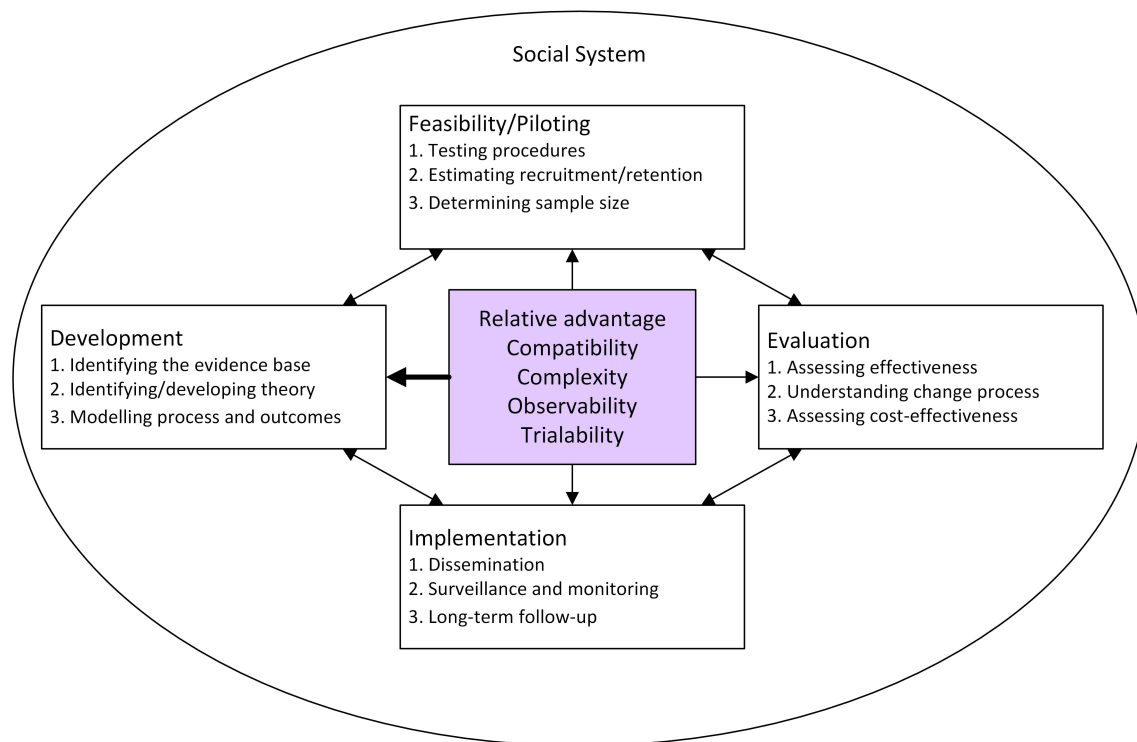
#### **7.3.3.2 Characteristics of the Intervention**

When applying the Diffusion of Innovation theory to the development and evaluation of an ICT-based intervention using the UKMRC guidelines, intervention developers and researchers need to consider the unique characteristics of the intervention that will improve its adoption (and diffusion) in the intended population. According to Rogers (2003), these characteristics are the ICT-based intervention's relative advantage, compatibility, complexity, trialability, and observability. I propose that ICT-based interventions would be more readily adopted if developers and researchers, who are using the UKMRC guidelines, place the five characteristics of their intervention at the centre of their process (see Figure 7.4). While I recommend that these characteristics be considered at all phases of the process, I believe they would have greater importance in the development phase



(represented by the bold arrow in Figure 7.4).

The poor adherence and notable drop-out rates reported in Chapter 4, together with the partial support for the appropriateness of the TOBY app in Chapter 5, suggest that some characteristics of the TOBY app did not facilitate adoption by the parents and their children. Moreover, if the developers considered the characteristics with respect to the Diffusion of Innovation theory and addressed them earlier in the TOBY app's development phase, the result could have been a more feasible, effective and appropriate intervention.



**Figure 7.4 Innovation characteristics with the UKMRC framework**

### 7.3.3.3 Preventative Innovations

A unique approach for developers and clinicians who wish to accelerate the adoption of feasible, effective and appropriate ICT-based interventions could be to regard them as preventative innovations within the Diffusion of Innovation theory. Rogers (2002, p. 991) defines a preventative innovation as “...a new idea that requires action at one point in time

in order to avoid unwanted consequences at some future time”. Preventative innovations are harder to adopt and take more time to diffuse through social contexts when compared to other innovations. This is because the rewards of the innovation are delayed in time, are relatively intangible, and may not even occur (Rogers, 2002, 2003). Important to note is that the term preventative is not used in the context of “preventing ASD” or with the expectation that children will outgrow their ASD traits, but in the context of reducing the challenges children with ASD could face in the future through the development of adaptive skills at an early age. Based on the definition, I would consider ICT-based interventions delivering EIBI to possess some of the characteristics of a preventative innovation. That is, ICT-based interventions support the attainment of developmental milestones in the short-term, which, in turn, could increase the likelihood of meeting future milestones that may not have happened if the support was not given at the time. Given the challenges of poor treatment adherence and the notable drop-out rate I faced (Chapter 4) and partial support for the appropriateness of the TOBY app (Chapter 5), I certainly support the notion that ICT-based interventions for children with ASD possess the characteristics of a preventative innovation.

Rogers (2002) suggests the following five strategies to improve the diffusion of preventative innovations: 1) change the perceived attributes of preventative innovations; 2) utilise champions to promote preventative innovations; 3) change the norms of the system regarding preventative innovation through peer support; 4) use entertainment-education to promote preventative innovations; and 5) activate peer network to diffuse preventative innovations. Understanding ICT-based interventions from this unique perspective could provide some novel recommendations and strategies for developers and researchers when developing ICT-based interventions.

### **7.3.4 Recommendations**

Acknowledging the iterative nature of the UKMRC guidelines and with respect to the integrated Diffusion of Innovation theory (including preventative innovations) by Rogers (2003), the work by Dingfelder and Mandell (2011) and the findings from my research, in the section that follows I propose strategies for developers and researchers that may increase the likelihood of adoption and diffusion for ICT-based interventions for children with ASD in regional and remote Australia. The strategies are presented in the context of the individual phases of the UKMRC guidelines (Craig et al., 2013).

#### **7.3.4.1 Development phase**

Firstly, I recommend that ICT-based intervention developers and researchers facilitate co-production of the intervention by involving the end-users and communities in which the intervention is to be used, early in the development process. Working *with*, and not just *at* the consumers is a key mindset that all developers and researchers should incorporate when developing ICT-based interventions for children with ASD (Fletcher-Watson, 2015). While it is essential during the developmental phase to ascertain the evidence base, identify and develop theory, and model processes and outcomes, it is also important to develop an in-depth understanding of the characteristics of the intervention that will influence its ultimate adoption. Seeking out change agents and opinion leaders with established communication channels throughout the social system for which the intervention is intended is an important strategy to improve the likelihood of rapid adoption.

Additionally, there may be utility in conducting thorough scoping activities within the community to identify who these change agents or opinion leaders are. This step cannot be overemphasised, particularly in the context of regional and remote communities where a strong sense of community for children with ASD is reported, and hence strong

communication channels could be leveraged to accelerate the diffusion of the ICT-based intervention throughout the community (Hoogsteen & Woodgate, 2013). For families living in regional and remote areas, examples of change agents or opinion leaders could be clinicians, other parents, paediatricians, general practitioners and teachers within targeted communities or across multiple communities. Lastly, it is essential that the key outcomes the ICT-based intervention is aiming to achieve are considered salient to the end-user at this stage of the intervention development; recruiting key community partners and opinion leaders could be an effective strategy to help identify these outcomes for families living in regional and remote areas (Dingfelder & Mandell, 2011).

Another key element of the Diffusion of Innovation theory and the innovation-decision process are the communication channels (information exchange) between one individual and another (or many others). In the context of ICT-based interventions, findings from Chapter 2 and Chapter 5 suggest that regular contact with a therapist or health professional would improve intervention adherence. Furthermore, in the only paper that I am aware of that applies the Diffusion of Innovation theory to a intervention specifically designed for children with ASD (a telehealth-based parent-mediated intervention, called ImPACT Online), the authors stressed the importance of receiving therapist support to facilitate the diffusion of the intervention throughout the social system in their study (Pickard et al., 2016). Given the higher propensity for avoidance coping for parents living in regional and remote areas as reported in Chapter 3, regularly scheduled contact may help support parents should they experience any challenges during implementation.

Developers or researchers who are in the initial stages of developing an ICT-based intervention should consider incorporating a communication channel between end-users and clinicians. As previously discussed, videoconferencing is a proven modality that may

be beneficial for parents to communicate directly with therapists, particularly for those who live a considerable distance from therapists (Wainer & Ingersoll, 2015). This communication channel could operate at an individual level with direct one-on-one interactions or could disseminate mass communication where one therapist can communicate with multiple users at once. Integrated communication channels could go some way in reducing the complexity of the intervention for parents by providing them with the means to ask questions and troubleshoot with therapists.

In relation to the characteristics of the intervention, ICT-based interventions should not only be easy to use, but enjoyable to use for both parents and children alike. Parent responses in Chapter 5 suggested that the TOBY app did not provide enough customisation options, therefore limiting its capacity to be individualised to their child. Parents also reported a high level of importance of the app, but some experienced difficulty engaging their children. Improved customisation options within ICT-based interventions could result in parents having greater success in engaging their children. In relation to the TOBY app, an ability to take and upload photos of objects in their own homes that match daily living skill activities, and the option to choose the gender of the therapists providing instructions to the child within the solo activities were some suggestions made by parents to address these issues. The ability to customise ICT-based interventions is also supported in the literature, with a systematic review into computer-based interventions to improve social and emotional skills in children with ASD highlighting its importance (Ramdoss et al., 2012). Improving the customisation capacity of ICT-based interventions could also improve their relative advantage against its competitors, further increasing its chances of adoption.

Developers and researchers should concentrate their effort on reducing the complexity of

the intervention, making it easy and intuitive to use for both the parents and children. A strength of the TOBY app, as reported by parents in Chapter 5, was its relative ease of use. The TOBY app included instructional videos and comprehensive directions that all parents reported were easy to understand and follow. The reduction in complexity is even more salient for ICT-based interventions, as when asked to rate their overall technology skills on a scale of one to ten in Chapter 5, not all parents rated their skills highly (mean = 6.9 on a scale of 0 to 10).

Feedback from the parents should be sought very early on in the development phase of the ICT-based interventions, specifically to its complexity and ease of use. Moreover, if researchers are providing support to parents, then they should plan to dedicate adequate resources to train the parents in the use of the intervention. Lastly, family support groups, peer-support groups via online social media platforms, information accessible via a website, or telehealth communications could provide timely solutions to problems and accessible information to support parents using ICT-based interventions.

#### **7.3.4.2 Feasibility/Piloting Phase**

Similar to the development phase, throughout the feasibility phase of the UKMRC guidelines, researchers and developers should continue to seek feedback from the community, end-users and opinion leaders to ensure the treatment addresses their needs; maintaining the intended relative advantage and a suitable fit with the social system within which it will diffuse. Furthermore, a communication channel between the end-user and the developers or researchers will also allow for feedback from users that developers could incorporate before commencing with larger-scale studies. Researchers and developers should also have the required skills to collect, analyse and interpret qualitative data to obtain accurate and meaningful feedback from their end-users. This will enable researchers

and developers to interpret the feedback appropriately and make changes to their intervention that best serve the needs of their end-users.

The feasibility phase would also be an appropriate time to assess the ICT-based intervention's compatibility with existing systems at a smaller scale. As a result, when it is time to scale the intervention during the effectiveness and implementation phases, any compatibility issues have already been considered and addressed. Some examples of compatibility issues to consider would be parents' access to the internet or ICT devices (computers or tablets), convenient and easy to navigate licencing processes, parents' technology literacy, and whether the intervention could be incorporated into the education system by teachers or education assistants. Developers could provide the opportunity for opinion leaders or change agents, in addition to participants, to trial and experiment with the intervention during this phase to improve the trialability and observability of the intervention.

#### **7.3.4.3 Evaluation Phase**

During the evaluation phase, researchers should not only use validated outcome measures that address the effectiveness of the intervention, but also capture data that measures the five characteristics of the intervention that will influence its adoption. Potential outcomes to be considered may include recording: 1) the number of community partners willing to advertise or provide feedback about the study; 2) the number of participants who responded to recruitment but did not meet the inclusions or exclusion criteria, or inquired after the closure of recruitment; 3) the degree of technical issues faced and resources that needed to be allocated to resolve them; 4) end-user active engagement and other metadata of the intervention in an easily accessible and interpretable manner; 5) the number of drop-outs; and 6) data on staffing resources required to support the intervention implementation

(hours of work) and other associated implementation costs. This data will enable researchers to report on the likelihood of adoption, as well as the overall effectiveness of the ICT-based intervention.

In line with the findings in Chapter 5, the likelihood of diffusion would increase if multiple communication channels were used to provide ongoing support to parents implementing the intervention during the evaluation phase. Examples of these include facilitating the communication between families who are implementing ICT-based interventions and support from educators, disability support workers, and general practitioners through regular follow-up with parents. Creating a community of people who are using the ICT-based intervention at the same time may improve the experience for end-users, as well as treatment fidelity and observability. Peer support may also assist in managing expectations for parents who may be seeing slow changes or benefits in using the ICT-based intervention with their child. Thus, parents may be more motivated to persevere with the intervention for an extended period, before abandoning the intervention if their expectations are tempered.

#### **7.3.4.4 Implementation Phase**

Communication channels within ICT-based interventions could also be useful for developers to disseminate their findings, conduct surveillance and monitoring, and assess maintenance of skills in the long-term with their study participants. Developers and researchers could utilise other technologies, such as social media, to diffuse their ICT-based intervention more easily directly to the parents (or other intended end-users). This would link very closely with effective knowledge translation strategies using targeted media and other peer-to-peer networks.

Researchers could also leverage champions and key opinion leaders to increase the success



of dissemination activities. End-users, generally speaking, do not evaluate an innovation based on the findings from scientific studies, with most people making their decision to adopt an innovation based on the subjective evaluation of the innovation communicated to them by other individuals who have already adopted the innovation (Rogers, 2003).

Researchers should ensure their dissemination activities incorporate adequate knowledge translation components, thus leveraging this theory through the champions and key opinion leaders identified early in the process.

Moreover, when disseminating their work, developers and researchers should focus on the relative advantage their ICT-based intervention has over other alternatives and be mindful of their competitors. Sometimes creating awareness with their intended end-users can be quite challenging, given the high level of noise and number of competitors, particularly for tablet-based interventions. However, the use of effective knowledge translation strategies in combination with strategies to differentiate their ICT-based intervention from alternatives that do not have evidence of effectiveness will highlight to parents of children with ASD the relative advantage of one ICT-based intervention over another.

## **7.4 Future Research**

### **7.4.1 Negative impacts of technology**

Most of the literature frames ICT-based interventions from a positive perspective, with little research having been conducted into how ICT-based interventions, or ICT more broadly, can negatively impact children with ASD. When I explored parents' experiences implementing the TOBY app during the interviews, it became apparent that some parents experienced unforeseen adverse outcomes with their child while using ICT. The following response from one of the parents resonated with me:

*Well, it didn't work for us because my son gets too obsessed with technology and his behaviour and his abilities were going backwards from being on the iPad too much.*

I acknowledge the adverse outcomes could not be attributed directly to the TOBY app, but perhaps could be attributed more broadly to the use of ICT. However, based on my findings, further research into the negative impact of ICT on children with ASD is warranted, with a particular focus on daily usage and transitioning from the iPad to other activities. Based on my experience working with participants and similar trends reported in literature, it was apparent that children with ASD often have a high affinity for ICT, which is perhaps not limited to this population, but can be applied to children more broadly (Ploog, Scharf, Nelson, & Brooks, 2013; Rajendran, 2013). However, I hypothesise that the challenges parents of children with ASD face with transitioning away from ICT use and limiting the daily consumption of these technologies would have some unique population-specific characteristics and further investigation would be beneficial.

#### **7.4.2 Cost-effectiveness**

Demonstrating a robust cost-benefit argument for ICT-based interventions could accelerate adoption (and diffusion) through policy and funding opportunities. Furthermore, given ICT-based interventions' unique characteristics to provide EIBI in scalable, standardised and remote-delivered methods, economic modelling to compare the cost-benefit against more traditional models is warranted.

Economic analyses of ICT-based interventions should consider both the direct service level perspective and the broader societal level perspective (Byford et al., 2015; Horlin, Falkmer, Parsons, Albrecht, & Falkmer, 2014). For example, economic evaluations of direct services should incorporate costs associated with medical and therapy services, intervention

delivery, purchasing the technology required to implement it, and providing ongoing support. While a societal level perspective could consider the broader economic evaluation in relation to childcare costs and future educational costs, parent-related expenditure (e.g., training courses, mental health services), losses in productivity of caregivers, travel associated with accessing services, and informal (unpaid) care due to the challenges associated with ASD should also be considered. This type of economic analysis has been conducted for other interventions for children with ASD, but is yet to be undertaken for ICT-based interventions for children with ASD (Byford et al., 2015).

#### **7.4.3 What works for whom and why?**

A finding from the appropriateness study (Chapter 5) with the subtheme, '*The TOBY app is not for everyone*' is congruent with other studies investigating predictors for symptom change in children with ASD. Due to the highly heterogeneous nature of ASD, children with ASD respond very differently to the same interventions and more research is required to ascertain "What works for whom and why?" (Hudry et al., 2018; Vivanti, Prior, Williams, & Dissanayake, 2014). I was unable to conduct mediation analyses in my research to determine the factors that resulted in the best outcomes for both parents and children due to a lack of statistical power. Future research should use larger sample sizes and conduct mediation analyses to identify both parent and child characteristics that result in the best outcomes for the TOBY app (or other ICT-based interventions).

#### **7.5 Limitations**

Throughout my research journey, I learnt a tremendous amount through my mistakes, and like all research, my research has some limitations. Despite an a priori decision to recruit families living in remote and very remote areas of WA for all studies, families from these areas were underrepresented. In particular, to accurately capture the unique experiences

and characteristics of families from remote and very remote areas of WA in Chapter 3, the recruitment of a higher proportion of this population was needed.

In the RCT reported in Chapter 4, I experienced poor adherence and a notable number of drop-outs, thus limiting the number of participants that completed the prescribed intervention at the recommended dosage. The notable number of drop-outs and poor adherence could have been ameliorated with additional support from the researcher, as highlighted in Chapter 2. The decision to provide limited support was made to improve the ecological validity of the TOBY app for families living in regional and remote areas. That is, families living in regional and remote areas are more likely to have limited support and the level of support we provided in the research aimed to reflect the real-life situation for these families.

In hindsight, providing limited support was perhaps an ill-advised decision. Given the findings from Chapter 2 and other research, it would have been more prudent to incorporate support during the evaluation phase to increase the likelihood of treatment adherence and reduce drop-outs. The findings do, however, actively support the existing evidence that ongoing support to parents who live in regional and remote areas is a necessary aspect of ICT-based intervention delivery. Also worth noting is that the trial by Whitehouse et al. (2017) that provided fortnightly calls to their participants experienced a higher participant drop-out rate (23%) to what I experienced (15%). Hence, the negative impact of not making fortnightly calls in our study may be somewhat overstated.

Regardless of whether the notable drop-out rate could be attributed to reduced levels of support, it did restrict the statistical power of analyses. As a consequence, the study became more exploratory in nature, thereby limiting the level of evidence. To accommodate the high attrition during the RCT, I should have been more proactive in recruitment, ensuring

an over enrolment in the study. Despite this limitation, findings from Chapter 4 contributed to the knowledge base by reporting on the preliminary effectiveness of the TOBY app and highlighting implications for future research into ICT-based interventions.

Another challenge experienced in the RCT described in Chapter 4, was the recruitment of children aged two to six; an initial inclusion criterion for the study. All reasonable attempts were made including dissemination through existing research databases, health services, professional networks and support groups. Despite this, the inclusion criteria of the trial needed to be expanded from children with a chronological age of two to six to children with a developmental age of two to six. Difficulties in recruiting younger children could be attributed to delayed diagnoses as a result of living in regional areas; however, this hypothesis could not be substantiated with the data available. Regardless, children with a developmental age of between two and six years were recruited and this may have influenced their engagement, and therefore, the treatment adherence.

Finally, the effectiveness trial lacked parent outcome measures. The decision to limit parent outcome measures was informed by the systematic review highlighting a significant lack of child outcome measures and as a result of attempting to limit the burden on parents participating in the trial. Parent data could, however, have provided valuable insight into the overall evaluation of the TOBY app, such as measuring parent's knowledge and treatment fidelity on ABA strategies or other associated benefits of engaging with the TOBY app.

## **7.6 Conclusion**

Throughout my thesis, I applied the UKMRC guidelines for developing and evaluating complex interventions to investigate the feasibility, effectiveness and appropriateness of the TOBY app. The TOBY app is an ICT-based parent-delivered intervention on an iPad,

grounded in ABA principles, that aims to improve the performance of children with ASD living in regional and remote WA in the four major areas of visual motor skills, imitation skills, language skills, and social skills (Craig et al., 2013; Moore et al., 2015; Venkatesh et al., 2013). Children with ASD often experience difficulty in these areas, which are vital building blocks for engaging in meaningful play, communication and socialisation (Baron-Cohen, 1987; Jarrold, Boucher, & Smith, 1993; Stagnitti & Unsworth, 2000).

Firstly, I contextualised my research by conducting a systematic review into parent-mediated interventions delivered remotely to families of children with ASD living in regional and remote areas to scope the literature landscape for similar interventions (Chapter 2). Findings from Chapter 2 concluded that parent-mediated interventions for children living in regional and remote areas delivered remotely have some effect in improving treatment fidelity and increasing parents' knowledge in ASD. Further, training for parent-mediated interventions delivered using video was more effective and appropriate compared to written training. Regular contact by therapists resulted in improved program completion, intervention fidelity, parent engagement, intervention appropriateness, and an improved perception of the child by the parent when compared to self-directed (i.e., minimal therapist contact) programs. Lastly, the systematic review provided an overview of current parent training interventions and different delivery methods for children with ASD living in regional and remote areas.

Through the cross-sectional survey I identified that there are few differences between the stress, coping, QoL and daily routines of families of children with ASD aged 2-18 living in LDP areas with those living in more DP areas (Chapter 3). However, families of children with ASD in WA, regardless of whether they live in LDP areas or more DP areas, experience higher levels of stress and poorer QoL compared to the general population.

Therefore, parents living in LDP areas seem to be as equally equipped to deliver ICT-based interventions compared with their DP living counterparts.

Through the use of an RCT in Chapter 4, I identified that the TOBY app is effective in improving the expressive language skills of children in the experimental group. After combining the data from the waitlisted control group with the experimental group, my research also supported the TOBY app's effectiveness in improving receptive language skills, pragmatic language skills, and social communication skills over a three-month intervention period. However, it was not effective in improving the visual motor skills, fine motor skills, and playfulness of children in the study.

To date, no research had been conducted to determine if the TOBY app is appropriate for use in families with a child with ASD living in regional and remote WA. In Chapter 5, I interviewed parents from the RCT (Chapter 4) and applied rigorous qualitative methodology to generate themes from their responses. I then applied these themes to a theoretical framework to evaluate the TOBY app's appropriateness for families living in regional and remote WA (Chapter 5). The parents' collective responses reported the TOBY was relevant and important to them and their children's needs, and partially supported the TOBY app as an appropriate intervention, resulting in: 1) a positive experience for them and their children; 2) perceived benefits for them and their children; 3) a socially and ecological valid intervention; and 4) an intervention that supported change and continuation in the skills learnt. However, they reported the TOBY app is not for every child with ASD, requires ongoing support from therapists when being implemented (reinforced by findings in Chapter 2), and is just part of the therapy that children with ASD should receive. That is, the TOBY app cannot and should not replace face-to-face therapy for families living in regional and remote WA.

A long-term follow-up study was completed following the RCT (Chapter 6). This was the first study to assess if the skills learnt while using the TOBY app were maintained in the long-term. Participants who used the TOBY app for three months gained and maintained skills for up to at least 12 months for receptive language, social and symbolic communication, pragmatic language and playfulness.

The results from this research provide partial support for the TOBY app as a feasible, effective and appropriate intervention for families of children with a developmental age of two to six years with ASD in regional and remote WA.

Finally, I provided clinical implications and recommendations for future researchers, clinicians and ICT-based intervention developers to improve the feasibility, effectiveness, appropriateness and adoption of ICT-based interventions for families of children with ASD living in regional and remote areas. The application and integration of the Diffusion of Innovation theory to the adoption of the TOBY app for families of children with ASD living in regional WA, and more broadly, in ICT-based interventions for children with ASD, is novel. These recommendations have the potential to provide insight for other populations who may use ICT-based interventions, as long as due consideration to the unique social context is given.



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Every reasonable effort has been made to acknowledge the owners of the copyright material used in this thesis. The original authors of the questionnaires and models used were contacted and written approval was obtained for their use in the PhD research. I would be pleased to hear from any copyright owner who has been omitted or incorrectly acknowledged.

Signature: \_\_\_\_\_

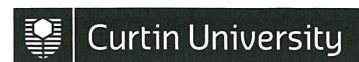
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September 17, 2019





# Appendix A Human Research Ethics Committee approval



## Memorandum

<b>To</b>	Professor Torbjorn Falkmer, School of Occupational Therapy and Social Work
<b>From</b>	Professor Peter O'Leary, Chair Human Research Ethics Committee
<b>Subject</b>	Protocol Approval <b>HR 123/2014</b>
<b>Date</b>	26 June 2014
<b>Copy</b>	Dr Sharmila Vaz School of Occupational Therapy and Social Work

Office of Research and Development  
**Human Research Ethics Committee**

**TELEPHONE** 9266 2784

**FACSIMILE** 9266 3793

**EMAIL** [hrec@curtin.edu.au](mailto:hrec@curtin.edu.au)

Thank you for providing the additional information for the project titled "*Autism in Rural Communities: The effect of an ASD diagnosis on rural families and innovative therapies*". The information you have provided has satisfactorily addressed the queries raised by the Committee. Your application is now **approved**.

- You have ethics clearance to undertake the research as stated in your proposal.
- The approval number for your project is **HR 123/2014**. Please quote this number in any future correspondence.
- Approval of this project is for a period of four years **01-07-2014** to **01-07-2018**.
- Your approval has the following conditions:
  - i) Annual progress reports on the project must be submitted to the Ethics Office.
- **It is your responsibility, as the researcher, to meet the conditions outlined above and to retain the necessary records demonstrating that these have been completed.**

### Applicants should note the following:

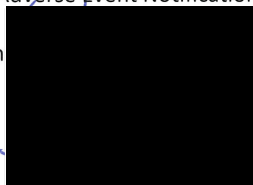
It is the policy of the HREC to conduct random audits on a percentage of approved projects. These audits may be conducted at any time after the project starts. In cases where the HREC considers that there may be a risk of adverse events, or where participants may be especially vulnerable, the HREC may request the chief investigator to provide an outcomes report, including information on follow-up of participants.

The attached **Progress Report** should be completed and returned to the Secretary, HREC, C/- Office of Research & Development annually.

Our website [https://research.curtin.edu.au/guides/ethics/non\\_low\\_risk\\_hrec\\_forms.cfm](https://research.curtin.edu.au/guides/ethics/non_low_risk_hrec_forms.cfm) contains all other relevant forms including:

- Completion Report (to be completed when a project has ceased)
- Amendment Request (to be completed at any time changes/amendments occur)
- Adverse Event Notification Form (If a serious or unexpected adverse event occurs)

Yours sincerely



Professor Peter O'Leary  
Chair Human Research Ethics Committee

# Appendix B Information Sheets and Consent Forms

## B.1 Survey

### B.1.1 Information sheet

## **Autism in Western Australia: Surveying the experience of families**

### **INFORMATION SHEET**

The Curtin Autism Research Group, in collaboration with the Disability Services Commission is conducting a survey to learn about the experiences of families with a child/young person (aged 2-18 years old) with Autism Spectrum Disorders (ASD). This survey is particularly timely with the role-out of the new National Disability Insurance Scheme (NDIS). It is also one of the most comprehensive surveys ever to be undertaken with families living with a child with ASD in Western Australia. Information collected from this survey will inform future services and policy. Importantly, this is the first wave of a longitudinal study that will allow us to learn how the needs of families with a child with ASD change over time.

#### **\*\*Who can participate in the survey?\*\***

If you are a parent or a primary caregiver of a 2-18 year old child/young person diagnosed with ASD, and live in Western Australia, you are eligible to participate in the survey.

#### **\*\*What will you be asked to do?\*\***

You will be asked to complete a survey on the processes families go through to obtain an ASD diagnosis and to access services, the additional costs associated with having a child with ASD, and the impacts of having a child/young person with ASD on the family's everyday life. The survey can be completed online, by telephone or paper and pencil. It takes approximately 60 minutes to complete the survey.

You will be given the choice to provide us with your personal details so that we can contact you in the future. You can also participate in the survey and choose NOT to provide us with your personal details.

You will be given the choice to give us permission to access your child's medical records and other Department of Health and Education registers. This will allow us to examine which treatments and investigations your child received and when they occurred.

#### **\*\*Does my family have to participate in the survey? \*\***

Participation in this survey is entirely voluntary, and you are free to withdraw at any time without providing a reason to do so.



The questions in the survey are designed to capture the experiences of ALL families with an ASD child/young adult in WA. We acknowledge that some sections of the survey may not reflect the personal experiences of some families. Some families have adjusted well following the diagnosis of their child with ASD, while other families are finding it difficult to adjust. The questions in the survey are not designed with one family in mind, but rather to capture the experiences of all families on the continuum of adjustment and acceptance. This means that some questions may not reflect your family's personal experience. If you feel uncomfortable in completing a particular section of the survey, you may continue to the next page by clicking on the *Next page* button on the bottom of the page.

**\*\*How do I access the survey? \*\***

**\*Please click the following link to access the survey:** <http://goo.gl/GmsIVR>

If you would like a **pencil and paper version** of the survey sent to you or if you have any further questions please do not hesitate to contact us either via e-mail on [autismstudy@curtin.edu.au](mailto:autismstudy@curtin.edu.au) or via the telephone on 0481 516 324.

Alternatively, you may directly contact the primary investigators listed below:

		
<p>Dr. Sharmila Vaz, Postdoctoral Research Fellow School of Occupational Therapy and Social Work Phone: 08-9266-1849 Email: <a href="mailto:s.vaz@curtin.edu.au">s.vaz@curtin.edu.au</a></p>	<p>Professor Torbjörn Falkmer, School of Occupational Therapy and Social Work Phone: 08-9266-9051 Email: <a href="mailto:t.falkmer@curtin.edu.au">t.falkmer@curtin.edu.au</a></p>	<p>Associate Professor Reinie Cordier, School of Occupational Therapy and Social Work Phone: 08-9266-2583 Email: <a href="mailto:reinie.cordier@curtin.edu.au">reinie.cordier@curtin.edu.au</a></p>

**B.1.2                      Consent form**

**Autism in Western Australia:  
Surveying the experience of families of children  
with Autism Spectrum Disorders (ASD)**

**Consent to Participate**  
(To be returned to the researchers)

**Prior to beginning the survey, we need to assess your eligibility.**

**Are you a parent/caregiver of a child aged 2 – 18 with any of the following diagnoses?**

Autistic Disorder (Autism), High-functioning Autism, Asperger's Syndrome, Pervasive Developmental Disorder – not otherwise specified (PDD-NOS), Rett's Syndrome or Childhood Disintegrative Disorder?

- ☐ Yes
- ☐ No

**Do you live in Western Australia?**

- ☐ Yes
- ☐ No

**What is your postcode?** \_\_\_\_\_

**Do you consent to participate in the current study?**

- ☐ Yes
- ☐ No

**Are you interested in participating in follow-up versions of this study?**

- ☐ Yes
- ☐ No

**Do you give the researchers permission to access your child's hospital records and other Department of Health and Education registers?**

- ☐ Yes
- ☐ No

**Would you like to be provided with information/be invited to participate in research projects in the future?**

- ☐ Yes  
☐ No

**This survey is the first part of a longitudinal study. To enable us to call you to complete the survey and follow up with you in the future, please provide us with your personal details. Your contact details will be removed from your responses and stored in a secure server that is only accessible to the researchers:**

**Please provide us with your contact details:**

Name (first and surname) \_\_\_\_\_

Address \_\_\_\_\_

City \_\_\_\_\_

State \_\_\_\_\_

Post Code \_\_\_\_\_

Email address \_\_\_\_\_

Phone number \_\_\_\_\_

Your date of birth [dd/mm/yy]\_\_\_\_\_

**Please return this copy of the consent form to.**

**Curtin Autism Research Group**

**Curtin University**

**Reply Paid 1987**

**Perth WA 6845**

## **B.2 Randomised controlled trial**

### **B.2.1 Parent and carer information sheet**

#### **iPad App Trial: Parent information sheet**

**The iPad intervention study in rural Western Australia: Examining an iPad App's effectiveness as a complementary therapy for 2-7 year olds recently diagnosed with Autism Spectrum Disorder (ASD).**

#### **What is this study about?**

This study has two aims:

1. To determine whether an iPad App is an effective complementary therapy for 2-7 year old children, recently diagnosed with ASD (or on the waitlist for an Autism assessment), who live in rural WA.
2. To examine parent/carers attitudes and beliefs towards the App as an intervention.

#### **Why has my family been contacted?**

You and your family have been approached because you have a child who has recently been diagnosed with ASD (or on the waitlist for an Autism assessment); and reside in the rural regions of the South West, Greater Southern, Wheatbelt, and Goldfields-Esperance.

#### **What will you be asked to do?**

**Telephone interview (10 minutes):** A member of the research team will contact you to explain the study and determine your child's eligibility to participate. We will ask questions about your child's age, diagnosis and medical condition. If your child is eligible, we will arrange a face-to-face meeting.

**Face-to-face meeting (2 to 3 hours):** This meeting can be conducted in your own home or at a pre-arranged venue convenient to you. Both you and your child will need to be in attendance. We will further explain the study requirements, answer your queries, and if you agree to proceed with the trial, we will ask you to sign a consent form.

After you sign the consent form, we will assess your child's developmental level by participation in fun play and child-friendly games. All of the assessment sessions throughout the study will be videoed for analysis purposes. Videos will be stored securely and only the researchers will have access to them. We will give you a home survey pack about your child's abilities and your family.

We will randomly allocate your child to one of two trial groups. The random allocation to one of the two groups will be generated by a computer.

- (i) iPad app group: Children in this group will be provided with an iPad (that they are able to keep) with the iPad app installed. Parents will be required to take part in an interactive information session providing training on the effective

use of the iPad App. Once again this can be conducted in your own home if desired. For the length of the trial, you will be asked to complete at least 20 minutes of therapy from the App each day for 3-months. This will be in addition to any other therapy the child is already undergoing outside of this study.

- (ii) ‘Therapy as Usual’ group: Children in this group will not be required to undertake any activities on the iPad App on top of whatever therapy they are already receiving outside of this trial - for the first 3 months into the trial. They will however be provided with an iPad that does not have the App installed and permitted to access different Apps for their child if desired. This may also give us an idea of how other Apps compare with the one we are evaluating.

The ‘intervention group’ will use the iPad App for 3 months. They will then become the “Therapy as usual” group and the participants in the original ‘Therapy as Usual’ group will become the “intervention group” for the next three months. Children in both groups will be assessed at entry into the trial (baseline) and at three and six-month follow-up time points. Children allocated to the original “therapy as usual” will have an additional follow-up at 9-months. The follow-up appointments will be arranged with you from the day you start the trial, typically at your child’s first assessment appointment.

**\*Follow-up 1 at 3-months – child assessment (1 hour)**

We wish to further assess your child three months after s/he enters the trial. We will make contact about two weeks prior to this meeting to remind you of the appointment. At this assessment we will examine your child’s development, using child friendly tasks and games. These assessments will be similar to those that your child had at entry into the trial. You will also be given a home survey pack

**\* Follow-up 2 at 6 months – child assessment (1-2 hours)**

Again, we wish to see your child six months after their entry into the trial (so, three months after follow-up 1 assessment). Similarly, we will make contact about two weeks prior to this meeting to remind you of the appointment. At this assessment, we will examine your child’s development, using child friendly tasks and games. For children allocated to the playpad group at the initial appointment, this will be the final appointment.

**\*Follow-up 3 at 9-months – child assessment (1 hour)**

If allocated to the “Therapy as Usual” group at your initial appointment, this will be your final appointment. We will make contact about two weeks prior to this meeting to remind you of the appointment. At this assessment, we will examine your child’s development, using child friendly tasks and games.

**Will my child miss out on therapy by participating in this study?**

No, we encourage you to continue to enrol your children in therapy services outside of this trial. At the time of entering into this trial, you will be provided with the contact details of Autism early intervention services in your local community, who are providing the best clinical practice for children with ASD.

**What are the benefits of participating in this study?**

At the conclusion of the study, **all families who take part in this trial (i.e., children in either group), will be able to keep the iPad provided and a copy of the iPad App.** The results from this trial may help to establish a new therapy method for delivering an early intervention for children with ASD. Your participation will contribute to this significant scientific advance.

**What are the disadvantages of participating in this study?**

This research involves time, effort and commitment from you and your child. You will be asked to meet with someone of the research team on three-four separate occasions over a nine-month period. Each appointment will be between one to three hours in duration. Among the battery of questionnaires, some items will ask you about personal and possibly deemed sensitive information about your family. You are at liberty to answer as many of the questions as you feel comfortable with. Importantly, all information is kept strictly confidential and secure.

**How and where will the information be stored?**

Any information that we collect from your family will remain private and confidential. The information gathered will be de-identified, which means that we will remove your names and give the information a unique code number. Only few of the research team members will be able to match your names to this unique code number, but only if it is necessary to do so. All written information will be stored securely in a locked filing cabinet at Curtin University, Perth. Electronic information will be stored on a password-protected computer database at Curtin University, Perth. Your child's name or any other identifiable information will not appear on any reports that may arise from this research.

**When will my family be informed of the results of this study?**

At the end of each year, you will receive a newsletter from Val Lishman Health Research Foundation that will inform you about the progress of this trial and other autism research studies taking place in our group. We are unlikely to be able to inform you of the findings from this research until the conclusion of the study.

**Does my family have to participate in the study?**

No. Participation in this study is entirely voluntary. You are free to withdraw your consent to participate in this study at any time. Your decision to withdraw consent will not influence your child's clinical management. All decisions made by your family will be respected by all members of the study team.



**Who is funding this study?**

This study is funded by Val Lishman Health Research Foundation.

**Who are the study investigators?****Chief Investigators**

Professor Torbjörn Falkmer

Dr. Sharmila Vaz

Associate Professor Reinie Cordier

Mr. Dave Parsons

Ms. Belinda Cuomo

**Does this study have ethical approval?**

This research has been reviewed and given approval by the Curtin University Human Research Ethics Committee (approval number HR123/2014). Should you wish to make a complaint on ethical grounds, please contact the Human Ethics Committee (Secretary), phone: 9266 2784, email: [hrec@curtin.edu.au](mailto:hrec@curtin.edu.au), mail: c/- Office of Research and Development, Curtin University of Technology, GPO Box U1987, Perth WA 6845. Your concerns will be drawn to the attention of the Ethics Committee who is monitoring the study.

**Who do I contact for more information about this study?**

If you would like to discuss anything about this trial, or get more information please contact Ms. Janine Slaven on 0497 549 773 or [Janine.slaven@curtin.edu.au](mailto:Janine.slaven@curtin.edu.au); or Mr. Dave Parsons on (08) 9266 3790 or [dave.parsons@curtin.edu.au](mailto:dave.parsons@curtin.edu.au); or Dr. Sharmila Vaz on (08) 9266 1849 or [s.vaz@curtin.edu.au](mailto:s.vaz@curtin.edu.au)

**What do I do to take part in this study?**

If you wish to take part in this trial, please:

Contact Ms. Belinda Cuomo on 0497 549 773 or [belinda.cuomo@curtin.edu.au](mailto:belinda.cuomo@curtin.edu.au); or Mr. Dave Parsons on (08) 9266 3790 or [dave.parsons@curtin.edu.au](mailto:dave.parsons@curtin.edu.au); or Dr. Sharmila Vaz on (08) 9266 1849 or [s.vaz@curtin.edu.au](mailto:s.vaz@curtin.edu.au)

**WE SINCERELY THANK YOU FOR YOUR CONSIDERATION  
WE LOOK FORWARD TO HEARING FROM YOU SOON.**

## B.2.2 Consent form

### iPad App trial: Consent form

#### **The iPad App intervention study in rural Western Australia: Examining its effectiveness as a complementary therapy for 2-7 year olds recently diagnosed with Autism Spectrum Disorder?**

*(Please note that participation in research studies is voluntary and subjects can withdraw at any time with no impact on current or future care)*

I, \_\_\_\_\_

(Given name)

(Surname)

- ☐ Have read the information explaining the study entitled “The iPad App intervention study in rural Western Australia: Examining its effectiveness as a complementary therapy for 2-7 year olds recently diagnosed with Autism Spectrum Disorder?”
  - ☐ Have understood the information given to me
  - ☐ Have had any questions asked answered to my satisfaction
- I agree to allow:

\_\_\_\_\_, my child, to participate in the study.

(Full name of participant)

- ☐ I understand my child may withdraw from the study at any stage and withdrawal will not influence his/her ongoing clinical care.
- ☐ I agree that if I withdraw from the study I am required to return the iPad to the research team in reasonable condition.
- ☐ I agree to replace the iPad if I withdraw from the study and the iPad is not in a reasonable condition.
- ☐ I agree to allow my child to undergo behavioural assessments at three, six and nine months after s/he is enrolled in the trial.
- ☐ I agree for the research information from the study to be published, provided that our names are not used.
- ☐ I understand that I might be allocated to the “iPad App control group” at the onset of the trial or after the first 3 months of the trial where I will not use the App being evaluated.
- ☐ I agree I have been given an iPad and I will place this iPad on my Home and Contents Insurance policy.

Date: \_\_\_\_\_ day of \_\_\_\_\_ 20\_\_

Parent or guardian’s signature: \_\_\_\_\_

*All research participants are entitled to keep a copy of the parent information sheet/ and participant consent form for their own records.*

### **B.3 Appropriateness study**

#### **B.3.1 Information sheet**

##### **TOBY Playpad iPad application:**

**An investigation to understand the parental experience of using TOBY-PA as a complementary therapy for families who have a child with ASD.**

<b>Participant Information Form</b>
-------------------------------------

##### **What is this study about?**

This study is seeking to explore your experience of using TOBY with your child. It is a follow-up study to the TOBY trial you have recently participated in. We are interested in what you feel made the application either difficult or easy to use and your opinion regarding what might improve TOBY.

##### **We invite you to participate**

As you are the parent/guardian of a child with an autism spectrum disorder, and you were randomly selected to be a part of the 'TOBY group' during the TOBY trial, you are eligible to be a part of this study. Participation in this study is voluntary.

##### **What will you be asked to do?**

Should you choose to take part in this follow-up study, we ask that you complete the attached expression of interest form and email a scanned copy directly to the research team at Curtin University conducting this follow-up study. Your form will be received by Dave Parsons, a PhD student at Curtin University. Dave will then contact you via your preferred method, to tell you if you have been selected from the respondents to take part in this follow-up study.

If selected, Dave will arrange with you a time and place of your choosing to be interviewed. This interview will be conducted with you by Dave in a private setting or via teleconference, whichever is more convenient. It is expected to take approximately one hour. Questions will be centered on your experience of using TOBY with your child, in addition to a few questions about your previous experience using iPad technology and your intentions to continue or discontinue use of TOBY in the future. The interview will be video recorded, so that we can recall your responses.

##### **What might be the benefit?**

Your opinion is important to us. We would like to know what it was about the TOBY application that you liked, or did not like, so that we can use this information to improve future versions of TOBY and other similar applications.

**Are there any risks to me?**

There are no known risks to participating in this follow-up study. Your involvement is completely voluntary. You may decide to withdraw at anytime without having to provide a reason, at which point you may ask that the information that you have supplied be destroyed.

If during the interview you do not wish to answer a question, you are welcome to choose not to respond without having to provide an explanation. You are also free to take a break at anytime during the interview or to reschedule the interview for another day. In the unlikely situation of your experiencing distress from the interview, the contact details of the lead researcher at Curtin University will be provided and we will encourage you to seek assistance from available support services such as your local GP, local counseling services like Relationships Australia (1300 364 277) or Lifeline (13 11 14).

**Who is the Curtin University research team?**

The research team conducting this study is headed by Professor Torbjorn Falkmer, a senior researcher at Curtin University.

Also contributing to the study is:

- Associate Professor Reinie Cordier, a researcher at Curtin University
- Dr. Sharmila Vaz, a researcher at Curtin University
- Mr. Dave Parsons, a lecturer and PHD candidate at Curtin University
- Ms. Janine Slaven, a research assistant with Curtin University

**How and where will the information collected be stored?**

All of the information we collect from you will remain private and confidential. The original recording of your interview will be deleted once transcribed, and in the transcription we will change your name and remove all identifying personal details. You will be provided the opportunity to select the name that replaces yours. A digital document linking your chosen replacement name to your actual name will be securely stored on Curtin University's Research Data Storage Drive with access only available to the Curtin University research team. We retain this information so that we can trace what contributions you made if necessary (e.g. If you decide to withdraw from the study and ask that we delete all of the information you provided, we will be able to sort your information from that of other participants).

The information that you provide will only be published in its de-identified form as scientific articles or may be presented at relevant conferences. All data will be uploaded to Curtin University's Research Data Storage Drive with access only available to the Curtin research team. We will not disclose any of your information to other parties except with your permission or if required to by law.

**Is this study approved?**

“Approval to conduct this research has been provided by the Curtin University, in accordance with its ethics review and approval procedures. Any person considering participation in this research project, or agreeing to participate, may raise any questions or issues with the researchers at any time.

In addition, any person not satisfied with the response of researchers may raise ethics issues or concerns, and may make any complaints about this research project by contacting the Human Ethics Committee (Secretary), phone: 9266 2784, email: [hrec@curtin.edu.au](mailto:hrec@curtin.edu.au), mail: c/- Office of Research and Development, Curtin University of Technology, GPO Box U1987, Perth WA 6845. Your concerns will be drawn to the attention of the Ethics Committee who is monitoring the study. All research participants are entitled to retain a copy of any Participant Information Form and/or Participant Consent Form relating to this research project.”

**Further Information**

If you have any questions or would like some more information about the study, please directly contact Dave Parsons at [dave.parsons@curtin.edu.au](mailto:dave.parsons@curtin.edu.au)

Alternately you may contact another member of the Curtin University research team:

- Dr. Sharmila Vaz:  
[s.vaz@curtin.edu.au](mailto:s.vaz@curtin.edu.au) or 9266 1849  
or
- Professor Torbjorn Falmker:  
[T.Falkmer@curtin.edu.au](mailto:T.Falkmer@curtin.edu.au) or 9266 9051

**Participation**

If you would like to be a part of this follow-up study please fill in the attached expression of interest form. Scan the completed form and email it to Dave at: [dave.parsons@curtin.edu.au](mailto:dave.parsons@curtin.edu.au)

If you do not wish to participate in this follow-up study, simple do not reply to this email and do not fill in the expression of interest form. Choosing not to participate in this follow-up study in no way affects your involvement in the original TOBY Trial.

**Thank you for your time and consideration.**

### B.3.2 Consent form

#### **TOBY Playpad iPad application:**

**An investigation to understand the parental experience of using TOBY-PA as a complementary therapy for families who have a child with ASD.**

#### **Consent to Participate (Participant Copy)**

(Please note that participation in this research study is voluntary and subjects can withdraw at any time with no impact on their involvement in the original TOBY Trial)

I, ..... ,  
(Given Names) (Surname)

- ☐ Have read and understand the information provided to me in the participant information form
- ☐ Agree to participate in this study as outlined to me
- ☐ Have been informed of and understand the purpose of this study
- ☐ Am aware that I can ask questions about this research, and I have been provided with the researchers' contact details
- ☐ Understand that there are no known risks in this study
- ☐ Understand that participation in this study is voluntary and that I can withdraw at any time without reason or consequence
- ☐ Have been informed that my personal information will be kept confidential and any identifiable information will not be used in published material
- ☐ Consent to this interview being video recorded and transcribed
- ☐ Consent to the researchers viewing my Autism Research Case History Questionnaire (previously completed for the PlayPad trial)
- ☐ Agree that the information I provide can be used in other studies and/or publications

Dated: ..... day of ..... 20 .....

Signature: .....

*"Approval to conduct this research has been provided Curtin University, in accordance with its ethics review and approval procedures. Any person considering participation in this research project, or agreeing to participate, may raise any questions or issues with the researchers at any time.*

*In addition, any person not satisfied with the response of researchers may raise ethics issues or concerns, and may make any complaints about this research project by contacting Human Ethics Committee (Secretary), phone: 9266 2784, email: hrec@curtin.edu.au, mail: c/- Office of Research and Development, Curtin University of Technology, GPO Box U1987, Perth WA 6845. Your concerns will be drawn to the attention of the Ethics Committee who is monitoring the study.*

*All research participants are entitled to retain a copy of any Participant Information Form and/or Participant Consent Form relating to this research project."*

## **B.4 Maintenance study**

### **B.4.1 Information sheet**

<b>HREC Project Number:</b>	
<b>Project Title:</b>	Maintenance of skills following the use of the ‘Therapy Outcomes By You Playpad’ (TOBY-PA): an iPad Application for children with Autism Spectrum Disorder.
<b>Principal Investigator:</b>	Lecturer, Dave Parsons
<b>Student researcher:</b>	Catherine Robinson
<b>Version Number:</b>	1
<b>Version Date:</b>	05/OCT/2016

#### **What is the Project About?**

Families of children with autism spectrum disorder (ASD) who live in regional Australia have limited access to early intervention from health services. Children with ASD often experience difficulty with social interactions and repetitive behaviours. Early intervention can be used to enhance their development, and teach social and emotional skills. However for early intervention to be effective the child needs to be engaged for at least 25 hours a week. This amount of therapy involves significant costs, and travel for people living in regional areas. Therefore therapies that utilise technology, such as iPad applications are gaining popularity as they can increase the hours of therapy without travel, and other associated costs. The ‘Therapy Outcomes By You Playpad’ (TOBY-PA) is an iPad application aimed at teaching children with ASD, social and behavioural skills. It is easily accessible from the App store for \$29.99, and can be used in the family home, without any input from health professionals. Studies have shown that the TOBY-PA may be effective in teaching children behavioural and language skills however it is not known whether children retain these skills over time.

This research is being conducted because we don’t know if after using the TOBY-PA application children are able to retain the skills that they have learnt, and continue to use these skills.

The aim of this project is to find out if children with ASD can retain skills that were learnt using the TOBY play pad.

This research is important in the further development of the TOBY application and other applications aimed at teaching skills to children with ASD or other developmental disorders. If children with ASD retain the skills learnt, it could lead to a decrease in the severity of symptoms. This could therefore reduce the hours of therapy needed from health professionals and consequently decrease the significant financial impact on

families and the health system. Maintenance of social and behavioural skills learnt, could make engaging in school and play occupations less challenging for children with ASD. This research could also be beneficial for increasing the access to therapy for people living in rural Australia. If the TOBY-PA is successful at teaching skills that children retain, then this application could be used as an additional form of therapy. As the TOBY-PA is easily accessible and low cost it would increase the hours of therapy without the travel and significant cost associated with therapy from health professionals.

This is a follow up project and we intend to have 20 children and their parents participating in this study.

### **Who is doing the Research?**

Catherine Robinson, an Occupational Therapy student at Curtin University, is conducting this research for her honours project. The project is funded by a \$1000 grant from the School of Occupational Therapy and Social Work at Curtin University. There will be no costs to you, and you will not be paid for participating in this research.

### **Why am I being asked to take part and what will I have to do?**

You have been asked to take part in this research, as it is a follow-up of the study you previously participated in called “Autism in Rural Communities: The effect of an Autism Spectrum Disorder (ASD) diagnoses on rural families and innovative therapies”.

The study will take place in your home at a time that is convenient to you. We ask that you complete an interview with the researcher, either when we visit you or via a phone call at a later date. In this interview we will ask you questions about the use of the TOBY-PA, such as ‘how often do you and your child use the application’? We will also get you to complete two questionnaires prior to the visit, and we will collect these on visitation. Two different assessments will be completed with your child when we visit your home. At this time we will also video record you and your child playing together for 20 minutes. We expect the visit will be for approximately 2 hours, with the interview taking 30 minutes and the assessments taking 1.5 hours to complete.

The interview will be audio recorded so we can concentrate on what you have to say and not distract ourselves with taking notes. A full written copy of the recording will be made after the interview. The 20-minute play session will be video recorded, so people who are blinded to the study can complete assessments using the recording.



**Are there any benefits' to being in the research project?**

There may be no direct benefit to you or your child from participating in this research. However we hope the results of this project will:

- Allow for further development of the TOBY-PA
- Impact access to other therapy services for those living in regional and remote Western Australia.

**Are there any risks, side effects, discomforts or inconveniences from being in the research project?**

The only foreseeable risk of being involved in this study is that your child could become distressed when completing assessments. To minimise this risk we will have you present at all times, and refer to counselling services if needed. If your child becomes distressed we will cease the assessments, and you can withdraw from the study. During the study other risks or benefits may become apparent. If this happens we will explain this to you and how it may affect you. You may be asked to complete another consent form to show that you understand the new information that we have given you. Other than giving up approximately 2 hours of your time, we do not expect there to be any inconveniences associated with being involved in this study.

**Who will have access to my information?**

The information collected in this research will be re-identifiable. This means that any identifying information will be removed and replaced with a numerical code. Only the research team have access to the code to link back to your child's name. All information collected is treated as confidential and will be stored either on a locked computer or in a locked cabinet. Any information we collect will only be used in this project unless otherwise specified.

All electronic data including video footage will be stored in a password-protected computer at Curtin University. All hard copies of research material will be stored in a locked cabinet at Curtin University. This data will only be accessible to the student researcher and research supervisors, and in the event of an audit or investigation, staff from the Curtin University Office of Research and Development. This data will be stored for 25 years, and will then be destroyed.

You have the right to access your information in accordance with relevant privacy laws. Results from this research may be published in professional journals or presented at conferences. However you and your child will not be identified in any of the results.

**Will you tell me the results of the research?**

We will not send you any results from this research however we will direct you to any publications of the results.

**Do I have to take part in the research project?**

Participation in this research is completely voluntary. You do not have to agree to participate if you do not want to. If you agree to participate but change your mind, you may withdraw from the project at any time. If you withdraw, you do not have to give a reason, and your information will not be used in the research.

**What happens next and who can I contact about the research?**

If you would like to ask any questions or get any further information regarding this study please contact Miss Catherine Robinson on [catherine.e.robinson@student.curtin.edu.au](mailto:catherine.e.robinson@student.curtin.edu.au).

If you agree to participate in this research we will ask you to sign the consent form. Signing this form indicates that you understand what you have read in this form, and that you agree to be involved in the research project. By providing consent you agree to have your information used as described above. Please take your time and ask any questions you have before you decide to participate. You will be given a copy of this information and the consent form to keep.

Curtin University Human Research Ethics Committee (HREC) has approved this study (HREC number 123/2014). Should you wish to discuss the study with someone not directly involved, in particular, any matters concerning the conduct of the study or your rights as a participant, or you wish to make a confidential complaint, you may contact the Ethics Officer on (08) 9266 9223 or the Manager, Research Integrity on (08) 9266 7093 or email [hrec@curtin.edu.au](mailto:hrec@curtin.edu.au).

**CONSENT FORM**

<b>HREC Project Number:</b>	
<b>Project Title:</b>	Maintenance of skills following the use of the ‘Therapy Outcomes By You Playpad’ (TOBY-PA): an iPad Application for children with Autism Spectrum Disorder.
<b>Principal Investigator:</b>	Lecturer, Dave Parsons
<b>Student researcher:</b>	Catherine Robinson
<b>Version Number:</b>	1
<b>Version Date:</b>	05/OCT/2016

- I have read, the information statement version listed above and I understand its contents.
- I believe I understand the purpose, extent and possible risks of my involvement in this project.
- I voluntarily consent to take part in this research project.
- I consent to being audio-recorded
- I consent to be video-recorded
- I have had an opportunity to ask questions and I am satisfied with the answers I have received.
- I understand that this project has been approved by Curtin University Human Research Ethics Committee and will be carried out in line with the National Statement on Ethical Conduct in Human Research (2007).
- I understand I will receive a copy of this Information Statement and Consent Form.

Participant Name	
Participant Signature	
Date	

Declaration by researcher: I have supplied an Information Letter and Consent Form to the participant who has signed above, and believe that they understand the purpose, extent and possible risks of their involvement in this project.

Researcher Name	
Researcher Signature	
Date	

*Note: All parties signing the Consent Form must date their own signature*

# Appendix C Interview Schedules

## C.1 Appropriateness study interview schedule

<b>Parent Name:</b>	
<b>Participant Number:</b>	
<b>Interview Conducted by:</b>	
<b>Date and time of interview:</b>	

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### Introduction

- Explain that the purpose of the interview is to see how you found the intervention and how we can improve it
- Ask if it is OK to record the interview over the phone

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### Information about child to help interviewer

- Age
- Gender
- Attend community school or home-schooled

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### Information about parent to help interviewer

- Age
- Gender
- Qualification
- Number of children in care
- Marital Status

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### Context

- Prior to this trial, did your family own an iPad? If so, tell me about the family's use of the iPad each week.
- What can you tell me about your child with ASD and their use of the iPad? Prompts – how often, average time used each day, Apps used, what context were the Apps used (e.g. to help with skills, education, leisure) was it connected to internet, websites visited ... etc
- So, tell me a bit about what sparked your interest in this project and why you were interested in using the App ?

Notes

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### **Child's Experience**

- What can you tell me about your child's experience using the application?
- What did they like about it / What didn't they like about it?

Notes

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### **Parent's Experience**

- What can you tell me about your experiences using the application? What did you like about it / What didn't you like about it?

Notes

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### **Effectiveness - Child**

- I am interested in gaining your perspective on how effective the TOBY was.
- Do you think your child benefited from using the intervention? How, why, in what context?
- What did you notice about the App at home? Impact, use, effect on others
- At school? In the playground? Did the teachers notice any changes? (if relevant)

Notes

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### **Effectiveness - Adult**

- Do you think that you benefited from using the intervention? How, why?
- What do you do / think differently since using the application?
- Are you still using the strategies from the intervention?

Notes

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### **Ease of use**

- I am interested in gaining your perspective on what made TOBY easy or difficult to use. Could you please give me an overview of what it was like using the TOBY application?
- Was there anything that you found difficult about completing therapy tasks? (As needed)
- Was there anything about application that you feel made it easy to use? (as needed)

Notes

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### **Improvement**

- How do you think we could improve the application?
- Any changes to the application?

Notes

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### **Support**

- **What supports do you think you and your child would benefit from over time?**
- **Would you see a benefit from regular contact from therapist checking in? If yes, how regularly?**
- **Longer duration needed?**

**Notes**

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### **Future Use**

- **Are you planning on using TOBY in the future with your child?**  
**Why (or why not)?**
- **Would you recommend using TOBY to a friend?**  
**(If no) Why not?**  
**(If yes) What advice would you give them regarding using TOBY?**

**Notes**



Introduce concept of 10-point scale – we’re going to get you to rate some things on a 10-point scale, 10 being fantastic and 0 being not so great

Overall, how would you rate your skills with technology?

0-----5-----10

What would you give out of 10 for your child’s experience of the application?

0-----5-----10

What would you give out of 10 for your experience of the application?

0-----5-----10

What would you give out of 10 for the benefits to your child in using the application?

0-----5-----10

What would you give out of 10 for benefits to you in using the application?

0-----5-----10

What would you give out of 10 for the applications usability (how easy or hard was it to use the application) with 10 being very easy and 0 being very difficult.

0-----5-----10

**Anything else you would like to add? Thank you.**

## C.2 Maintenance study interview schedule

### Semi-Structured Interview Guide

The student researcher will conduct a semi-structured interview with the parent/s of the child with ASD. Estimated running time is 30 minutes.

#### Guide:

- Introduce self and give an explanation of why the interview is being conducted.
- Discuss confidentiality and gain consent to audio record the interview.
- Ensure the participant has signed the consent form.

#### Explain to the parent:

The main reason for this interview is to find out the reasons for continued or discontinued use of the TOBY application.

#### Questions

1. How long has it been since your child was last assessed?
2. Do you and your child still use the TOBY application? (Choose whether question 3 or 4 is appropriate to ask from the answer to this question)
  - Prompts that may be used:
    - How many times a week or minutes per day do you use the application on average?
    - What aspect of the application do you use more- Solo/partner/NET. Why do you use these more/Why do you not use the others?
    - Will you continue to use the application?
3. Why don't you use the application, (or why do you use it less)?
  - Prompts that may be used:
    - What don't you like about the TOBY? /Was there anything that made using the TOBY difficult?
    - Is there anything you did like about the TOBY?
    - How did it fit TOBY into your daily schedule?
    - How satisfied are you with the TOBY as an intervention tool?
    - Is there anything that would make using the TOBY easier?
4. Why have you continued to use the TOBY?
  - Prompts that may be used:
    - What do you like about TOBY?
    - What didn't you like about TOBY?
    - How satisfied are you with TOBY as an intervention tool?
    - How do you manage fitting in the 20 minutes of therapy a day?
    - Is there anything that would make using TOBY easier?
5. Do you think this application has benefited your child?
  - Since the last assessments have you seen any improvement in your

child's:

- Social behavioural skills
- Expressive and receptive language skills
- Fine motor skill

**Rate the following statements on the 10-point scale, by placing a cross on the line.**

0= strongly disagree

5= neutral

10= strongly agree

I think that the TOBY-PA is easy to use

0-----5-----10

0-----5-----10

I find it easy to fit in 20 minutes of therapy a day with the TOBY

0-----5-----10

0-----5-----10

I prefer the TOBY to other forms of therapy

0-----5-----10

0-----5-----10

### Summary

Are there any changes you would recommend for the TOBY?

Thankyou for your time, do you have anything else you would like to add?

# Appendix D Copyright clearance

## D.1 UKMRC Guidelines

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Number of charts/graphs/tables/figures	1
The requesting person/organization is:	David Parsons
Title or numeric reference of the portion(s)	Figure 1
Title of the article or chapter the portion is from	"Developing and evaluating complex interventions: the new Medical Research Council guidance."
Editor of portion(s)	N/A
Author of portion(s)	Craig, Peter Dieppe, Paul Macintyre, Sally Michie, Susan Nazareth, Irwin Petticrew, Mark
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## D.2 Journal Articles

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Format	Print, Electronic
Portion	chapter/article
The requesting person/organization is:	Dave Parsons
Title or numeric reference of the portion(s)	Parsons, D., Cordier, R., Lee, H., Falkmer, T., & Vaz, S. (2018). A randomised controlled trial of an information communication technology delivered intervention for children with autism spectrum disorder living in regional Australia. Parsons, D., Wilson, N. J., Vaz, S., Lee, H., & Cordier, R. (In Press). Appropriateness of the TOBY application, an iPad intervention for children with Autism Spectrum Disorder: A thematic approach. Journal of Autism and Developmental Disorders.
Title of the article or chapter the portion is from	Parsons, D., Cordier, R., Lee, H., Falkmer, T., & Vaz, S. (2018). A randomised controlled trial of an information communication technology delivered intervention for children with autism spectrum disorder living in regional Australia. Parsons, D., Wilson, N. J., Vaz, S., Lee, H., & Cordier, R. (In Press). Appropriateness of the TOBY application, an iPad intervention for children with Autism Spectrum Disorder: A thematic approach. Journal of Autism and Developmental Disorders.
Editor of portion(s)	N/A
Author of portion(s)	Dave Parsons
Volume of serial or monograph.	49
Issue, if republishing an article from a serial	2
Page range of the portion	
Publication date of portion	2018
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## Appendix E Author contribution statements

### E.1 Author Contribution Statement: Chapter 2

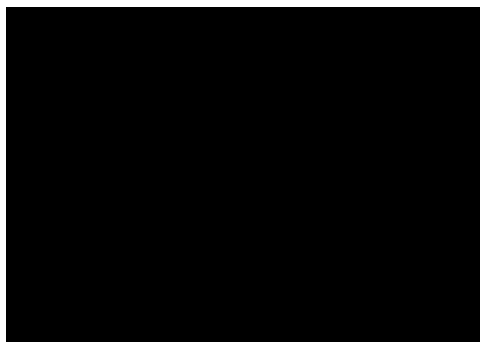
As co-authors of the paper entitled, “*Parent-mediated intervention training delivered remotely for children with Autism Spectrum Disorder living outside of urban areas: a systematic review.*” we confirm that Dave Parsons has been the principal researcher and has made the following contributions:

- Conceptualisation and design of the research;
- Data collection, analysis and interpretation;
- Writing the manuscript and critical appraisal of the findings;
- Corresponding author for communication with the journal

Our contribution to the paper was consistent with the role of supervisors and involved the following contributions:

- Assistance with conceptualisation and design of the research;
- Assistance with data analysis and interpretation; and
- Review and editing of the manuscript.

Signed



Reinie  
Cordier

Date:  
27/07/2019

Signed

Hoe Lee

Date: 25 July  
2019

Signed

Sharmila  
Vaz

Date: 7/8/2019

## E.2 Author Contribution Statement: Chapter 3

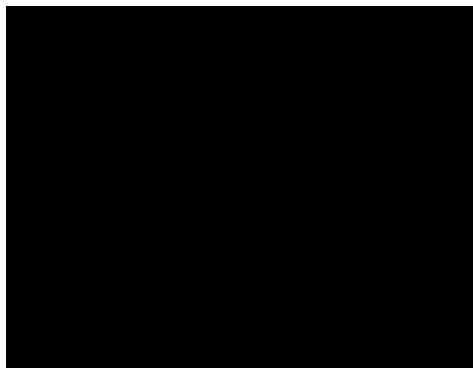
As co-authors of the paper entitled, “*Stress, coping, and quality of life in families with a child with ASD living regionally*”, we confirm that Dave Parsons has been the principal researcher and has made the following contributions:

- Conceptualisation and design of the research;
- Data collection, analysis and interpretation;
- Writing the manuscript and critical appraisal of the findings;
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- Review and editing of the manuscript.

Signed



Reinie  
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Date:  
27/07/2019

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Hoe Lee

Date: 25 July  
2019

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Sharmila  
Vaz

Date: 7/8/2019

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- Corresponding author for communication with the journal

Our contribution to the paper was consistent with the role of supervisors and involved the following contributions:

- Assistance with conceptualisation and design of the research;
- Assistance with data analysis and interpretation; and
- Review and editing of the manuscript.

Signed		Reinie Cordier	Date:
Signed		Hoe Lee	Date:
Signed		Sharmila Vaz	Date:
Signed		Torbjorn Falkmer	Date: 23/07/2019



### E.3 Author Contribution Statement: Chapter 4

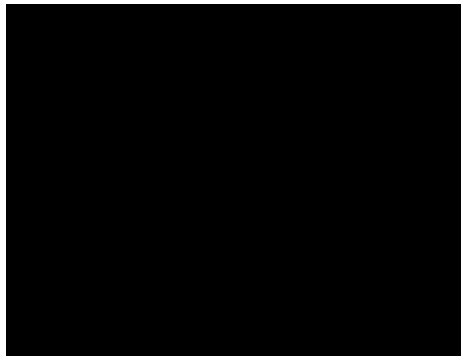
As co-authors of the paper entitled, “*A randomised controlled trial of an information communication technology delivered intervention for children with Autism Spectrum Disorder living in regional Australia.*” we confirm that Dave Parsons has been the principal researcher and has made the following contributions:

- Conceptualisation and design of the research;
- Data collection, analysis and interpretation;
- Writing the manuscript and critical appraisal of the findings;
- Corresponding author for communication with the journal

Our contribution to the paper was consistent with the role of supervisors and involved the following contributions:

- Assistance with conceptualisation and design of the research;
- Assistance with data analysis and interpretation; and
- Review and editing of the manuscript.

Signed



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Date:  
27/07/2019

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Hoe Lee

Date: 25 July  
2019

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Sharmila  
Vaz

Date: 7/8/2019

As co-authors of the paper entitled, "*A randomised controlled trial of an information communication technology delivered intervention for children with Autism Spectrum Disorder living in regional Australia.*" we confirm that Dave Parsons has been the principal researcher and has made the following contributions:

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- Assistance with conceptualisation and design of the research;
- Assistance with data analysis and interpretation; and
- Review and editing of the manuscript.

Signed		Reinie Cordier	Date:
Signed		Hoe Lee	Date:
Signed		Sharmila Vaz	Date:
Signed		Torbjorn Falkmer	Date: 25/07/2019

#### E.4 Author Contribution Statement: Chapter 5

As co-authors of the paper entitled, “*Appropriateness of the TOBY application, an iPad intervention for children with Autism Spectrum Disorder: A thematic approach.*” we confirm that Dave Parsons has been the principal researcher and has made the following contributions:

- Conceptualisation and design of the research;
- Data collection, analysis and interpretation;
- Writing the manuscript and critical appraisal of the findings;
- Corresponding author for communication with the journal

Our contribution to the paper was consistent with the role of supervisors and involved the following contributions:

- Assistance with conceptualisation and design of the research;
- Assistance with data analysis and interpretation; and
- Review and editing of the manuscript.

Signed



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Date: 27/07/2019

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Date: 25 July 2019

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Sharmila Vaz

Date: 7/8/2019

Signed

Nathan Wilson

Date: 21/08/2019

## E.5 Author Contribution Statement: Chapter 6

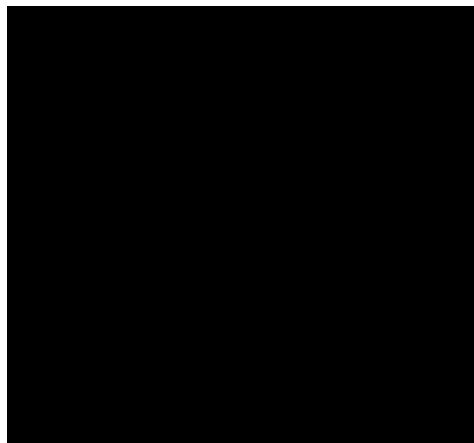
As co-authors of the paper entitled, “*A twelve-month follow-up of an information communication technology delivered intervention for children with autism spectrum disorder living in regional Australia.*” we confirm that Dave Parsons has been the principal researcher and has made the following contributions:

- Conceptualisation and design of the research;
- Data collection, analysis and interpretation;
- Writing the manuscript and critical appraisal of the findings;
- Corresponding author for communication with the journal

Our contribution to the paper was consistent with the role of supervisors and involved the following contributions:

- Assistance with conceptualisation and design of the research;
- Assistance with data analysis and interpretation; and
- Review and editing of the manuscript.

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Reinie  
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